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## Ileal lengthening through internal distraction: A novel procedure for ultrashort bowel syndrome

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### Abstract

**Purpose:** Ultrashort bowel syndrome is a rare, but morbid surgical problem without effective treatment. Recent clinical analysis has demonstrated the critical influence of ileal length on ultimate enteral autonomy. Surgical techniques to increase ileal length in nondilated bowel do not exist. We describe a novel technique to lengthen ileum in children with ultrashort bowel syndrome.

**Methods:** Beginning in May 2021 prospective candidate children were identified. Candidacy for ileal tube lengthening included diagnosis of ultrashort bowel syndrome, intact ileocecal valve with remnant ileum, and proximal intestinal stoma or draining gastrostomy. Informed consent was obtained. Following laparoscopic lysis of adhesions, a balloon catheter was inserted through a left flank stab incision and into the lumen of the remnant ileum around a purse string suture. Cecopexy was performed in the right-lower quadrant. Clips were used to mark the cecum and the proximal extent of ileum. The catheter length was fixed externally at the completion of the procedure. Serial x-rays were used to measure distraction effect while increasing tension was applied to the catheter over the subsequent weeks. Ileal tube lengthening was performed until the end of the catheter was reached or the tube was dislodged. A contrast study was performed at the completion

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CRediT authorship contribution statement

**Aaron J Cunningham:** Writing – original draft, Writing – review & editing, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Taylor Anderson:** Writing – original draft, Writing – review & editing, Visualization, Data curation, Conceptualization. **Claudia Mueller:** Writing – original draft, Writing – review & editing, Investigation, Data curation, Conceptualization. **Matias Bruzoni:** Writing – original draft, Writing – review & editing, Investigation, Data curation, Conceptualization. **James CY Dunn:** Writing – original draft, Writing – review & editing, Validation, Supervision, Resources, Methodology, Investigation, Formal analysis, Data curation, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

of lengthening. Intestinal length at time of restoration of continuity and clinical outcomes were recorded.

**Results:** Four infants were enrolled from May 2021-July 2023. Diagnoses leading to ultrashort bowel syndrome were mesenteric teratoma, necrotizing enterocolitis, and multiple intestinal atresia. At the time of restoration of intestinal continuity, a median of 1.75 cm (45 %) additional ileal length was achieved at a median of 25.5 days. There were no serious complications following ileal tube lengthening and no additional operative interventions were required.

**Conclusions:** Ileal lengthening through internal distraction is a feasible surgical intervention to salvage ileum for infants with ultrashort bowel syndrome. Ileal tube lengthening may result in distraction enterogenesis, providing a novel intervention to increase intestinal length.

**Level of evidence:** IV (Case series without comparison group).

### Keywords

Short bowel syndrome; Ultrashort bowel syndrome; Distraction enterogenesis; Intestinal failure; Intestinal lengthening

### Introduction

Short bowel syndrome (SBS) is a rare, but morbid surgical problem without effective treatment [1–4]. Many neonatal surgical conditions including necrotizing enterocolitis (NEC), congenital intestinal atresia, and midgut volvulus, among others result in loss of enteric mass and ultimately intestinal failure requiring long-term parenteral nutrition (PN) [5,6]. While the invention and use of PN has saved countless lives, long-term PN is associated with constant maintenance, recurrent hospital admissions, the potential of serious morbidity, and in some cases the inability to wean its long-term use [7–10]. Fortunately, the recent growth of intestinal rehabilitation programs has coalesced care and improved survival in children with SBS [11]. Nevertheless, few definitive therapies currently exist.

Notably in the 1980's with longitudinal intestinal lengthening (LILT) [12] and followed by serial transverse enteroplasty (STEP) in the early 2000's [13], surgical techniques have been developed to increase overall intestinal length as treatment for SBS. While these procedures have been successful at limiting overall PN use [14,15], they are principally used in the setting of significant bowel dilation and not commonly employed before six months of life [15,16]. Additionally, recent evidence would suggest that more than overall length, ileal length critically contributes to future enteral autonomy [17]. While both LILT and STEP have been successful in increasing overall length to treat SBS, they have not been commonly employed as treatment in the rare subset of ultrashort bowel syndrome (USBS), where small bowel length is less than 10 % of that expected for age [18]. In fact, use of LILT or STEP in these settings appears to be contraindicated because the key indicating factors of bacterial overgrowth or bowel dilation are absent.

Anatomically, children with USBS often have < 10 cm of intestinal length and commonly have a preserved remnant of terminal ileum (1–2 cm) with intact ileocecal valve. This specific anatomic situation – common in children with USBS – currently has no surgical

solution as they are inappropriate for LILT or STEP. Additionally, although children with USBS are known to be less likely than their counterparts with SBS to ultimately achieve enteral autonomy [19], there are currently no prophylactic interventions that exist for them and moreover, no surgical treatment specifically to lengthen ileum, which is critical to weaning PN.

Recently, the concept of distraction enterogenesis has been used to induce significant intestinal growth, notably in animal models [20–23]. Through mechanotransduction, intestinal distraction induces enterogenesis [24] leading to absolute growth in intestinal mass [25]. However, application has been limited to animals currently. To address these gaps, we describe a novel technique to lengthen ileum in children with ultrashort bowel syndrome and demonstrate its feasibility.

## Methods

### Data source and patient selection

Beginning in May 2021 prospective candidate children were identified from within the intestinal rehabilitation program at our institution. Candidacy for ileal tube lengthening included diagnosis of USBS, intact ileocecal valve with remnant ileum predicted to be at least 1 cm, and current enteric anatomy consistent with intestinal discontinuity. Successive parents were approached for ileal tube lengthening. The novelty of the procedure, risks, alternatives, and possible benefits were discussed and informed consent was obtained. The procedure and subsequent postoperative care were conducted as part of the clinical care of our intestinal rehabilitation program and not as part of an intended research study. As such, approval of the institutional review board was not obtained. Patient demographics, diagnosis leading to ultrashort bowel syndrome, preoperative ileal length, baseline intestinal anatomy, serial radiographic ileal length, post-lengthening ileal measurements, complications, and clinical outcomes were recorded.

### Definitions and outcomes

Ultrashort bowel syndrome was defined as the need for ongoing PN greater than 60 days after intestinal resection and less than 10 % of predicted intestinal length for age and size as defined by the North American Society for Pediatric Gastroenterology [1,26]. Preoperative ileal length was measured at the time of ileal tube placement, intra-operatively, while the ileum was under gentle traction, along the antimesenteric border. Radiographic ileal length was measured between the cecal clip and the lateral border of the enteric balloon or ileal clips. Post-lengthening ileum was measured at the time of restoration of intestinal continuity while the ileum was under gentle traction, along the antimesenteric border. Duration of lengthening was recorded from the time of ileal tube placement to end of distraction period.

### Procedure & lengthening

Following laparoscopic lysis of adhesions, a balloon catheter is inserted through a left flank stab incision and into the lumen of the remnant ileum. A purse string suture is used to tighten the intestinal lumen around the balloon and keep the catheter in place (Fig. 1). Cecopexy is performed in the right-lower quadrant with multiple interrupted

sutures to the abdominal sidewall. A single titanium clip is used to mark the cecum and two titanium clips mark the proximal extent of the ileum. The balloon catheter is filled with radio-opaque contrast (Omnipaque™ 350) so that the balloon could be visualized on subsequent radiographs. The catheter length is fixed externally with the ileum in a neutral position using a Biopatch™ and umbilical cord clamp over the exit site at the completion of the procedure (Fig. 2).

Tension is added to the ileal tube at discrete intervals by adding a Biopatch™ between the skin and the umbilical clamp on alternating days. Serial radiographs are used to measure distraction effect while increasing tension is applied to the catheter over the subsequent weeks (Fig. 3). Ileal tube lengthening is performed until the end of the catheter is reached or the tube is dislodged. When possible, a contrast study is performed at the completion of lengthening.

After lengthening, restoration of intestinal continuity is performed. Post-lengthening ileal length is measured (Fig. 4). Ileal-enteric anastomosis is performed in a single-layer interrupted fashion. Usual postoperative care is given until return of intestinal function.

### Statistical analysis

Intestinal length, time to events, and age are assumed to be nonparametric and are reported individually and as descriptive statistics with medians. Percent growth was calculated as relative growth from initial intraoperative measurements. Analyses were performed using IBM SPSS Statistics for Windows, version 25 (IBM Corp., Armonk, NY).

To determine rate of intestinal growth with ileal tube lengthening, aggregated radiographic measurements were plotted chronologically and linear regression was performed. Rate of growth and  $R^2$  are reported.

### Results

Four infants were enrolled from May 2021-July 2023. Diagnoses leading to ultrashort bowel syndrome were mesenteric teratoma, necrotizing enterocolitis, and multiple intestinal atresia (Table 1). Two patients were in complete enteric discontinuity with clips at the duodenum, a draining gastrostomy, and discontinuous inaccessible intra-abdominal enteric remnants. The remaining two patients had end jejunostomies with discontinuous inaccessible intra-abdominal enteric remnants. Median age at enrollment was 82 days, with a median preoperative ileal length of 4 cm.

Radiographic growth of the ileum was achieved in all patients with a median of an additional 3.08 cm achieved (Table 2). Median age at restoration of intestinal continuity was 132 days. At the time of restoration of intestinal continuity, a median of 1.75 cm or 45 % additional ileal length was achieved. Two patients developed superficial surgical site infections requiring extended duration antibiotics. No additional procedures or reoperations were needed after intestinal lengthening and restoration of intestinal continuity.

In regression analysis, radiographic ileal length is seen to increase at a rate of 5.9 % per day of lengthening (Fig. 5).

## Discussion

We report a case series of 4 children, demonstrating 1.75 cm and 45 % additional intestinal length over 1 month using an ileal tube technique. While radiographic growth appeared more considerable at 3.08 cm, after tension was released only 1.75 cm of growth was achieved. Nevertheless, the ileal tube lengthening procedure described is feasible and easy to implement. Lastly, through linear regression analysis, we demonstrate over 5 % growth per day with ileal tube lengthening. To our knowledge, this is the first report of successful distraction enterogenesis in humans.

With the persistence and prevalence of SBS, notably among children, there have been a number of attempts over the last 100 years to provide a surgical solution for intestinal insufficiency. Notably, the Bianchi procedure [12] and more recently the STEP procedure [13] have described surgical techniques to address intestinal stasis, dysmotility, and poor nutrient absorption associated with SBS [27]. These techniques rearrange the anatomic alignment of dilated and hypertrophied bowel but do little to increase the absorptive capacity of the intestine. Additionally, despite the use of LILT and STEP to treat dramatic bowel dilation, a long-term sequelae of these procedures is re-dilation in up to 57 % [28]. Furthermore, these techniques are not often used in the subgroup of children with USBS, and do not specifically address the ileum, which has recently been shown to be a critical contributor to ultimate enteral autonomy [17].

Distraction enterogenesis, alternatively, provides the promise of creating new intestinal mass through mechanotransduction and paracrine signaling [24,25]. Prior animal work has demonstrated significant growth of intestinal length (140–200 %) with distraction enterogenesis [20–22,29], but application in humans has been limited due to lack of a safe and suitable technique. With the description of the 4 preceding children, we demonstrate the feasibility of distraction enterogenesis in humans, with a focus on ileal growth. Notably, we report > 5 % increase in length per day which remarkably compares similarly to that demonstrated in porcine [5.25 % per day] [29], murine [4.29 % per day] [23], and rat models [6.7–10.5 % per day] [20,22].

There are no current standard measures to correlate length of ileum with future enteral autonomy, so the clinical importance of an additional 1.75 cm of ileum is unknown. However, ileal tube lengthening does provide a technique to allow for preservation of the ileocecal valve in the setting of a short terminal ileum segment. Surgical dogma would suggest that in settings where only a small segment of terminal ileum is preserved, the ileocecal valve be resected due to the theoretical risk of leak in an anastomosis so close to a functionally obstructed ileocecal valve. Despite the small size of the ileum in our study – ranging 2–8 cm post-lengthening – we report no anastomotic leaks after restoration of enteric continuity, both challenging surgical dogma and providing a solution to an age old surgical question. Future study will be required to determine the ultimate clinical significance of these findings and whether they lead to increase enteral autonomy. Further future areas of research could explore an attempt at re-lengthening the same segment, as it is clear that the bowel lengthens proportionally to its pre-existing size.

Ileal tube lengthening is a novel technique, specifically designed as a surgical treatment for the unique anatomic situation of near total intestinal loss, with a small remnant terminal ileum and intact ileocecal valve. Children with ultrashort bowel syndrome – especially those with near total intestinal loss due to NEC – not uncommonly have suffered ischemic injury to the majority of their ileum, but with preservation of the most distal remnant at the ileocecal valve. Ileal tube lengthening was designed specifically for these children, to maintain the valuable ileocecal valve and preserve the critical remnant ileum. We employed this technique exclusively in this circumstance as there is no current surgical treatment for this population.

We recognize a number of limitations to our study notably due to the small sample size of our population. Our case series is meant to demonstrate the feasibility of ileal tube lengthening and with no major complications and 1.75 cm gained, we propose that we achieve this goal despite the small sample. Further research is needed, however, to demonstrate safety given our small sample size. Care should be taken, however, in interpreting the results of our linear regression analysis, as a sample size of 4 may not sufficiently represent all intestinal growth rates. Additionally, the lack of long-term data and follow-up raise the question of efficacy of ileal tube lengthening, however in this study we provide the method for lengthening and propose that efficacy will be demonstrated in future studies. Lastly, as a single-center, single-surgeon study these outcomes may not be broadly applicable and further application and analysis are needed to demonstrate efficacy.

## Conclusion

Ileal lengthening through internal distraction is a feasible surgical intervention to salvage ileum for infants with ultrashort bowel syndrome. Through a straightforward procedure the ileum can be lengthened an additional 1.75 cm and 45 % using already available equipment. Ileal tube lengthening may result in distraction enterogenesis, providing a novel intervention to increase intestinal length in neonates.

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## Abbreviations:

<b>SBS</b>	short bowel syndrome
<b>USBS</b>	ultrashort bowel syndrome
<b>PN</b>	parenteral nutrition

<b>NEC</b>	necrotizing enterocolitis
<b>STEP</b>	serial transverse enteroplasty

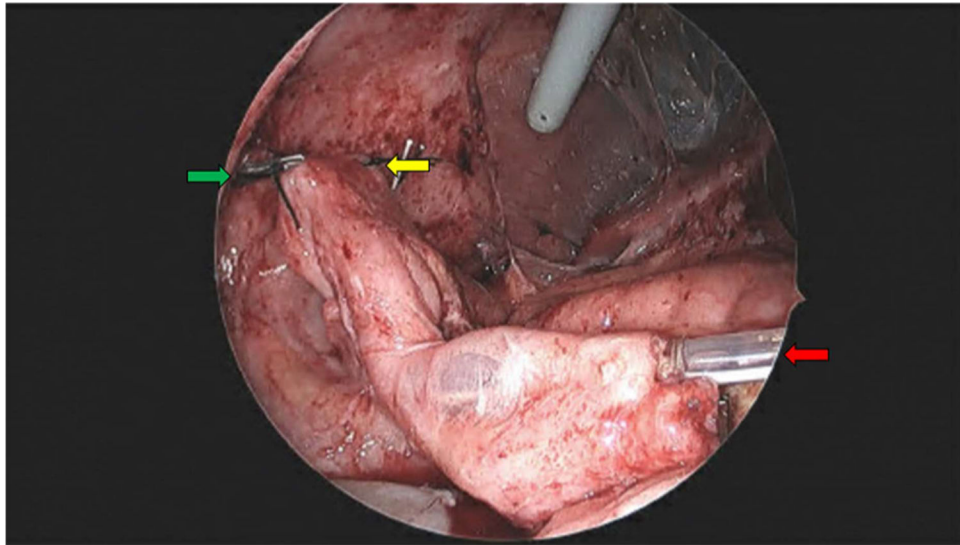
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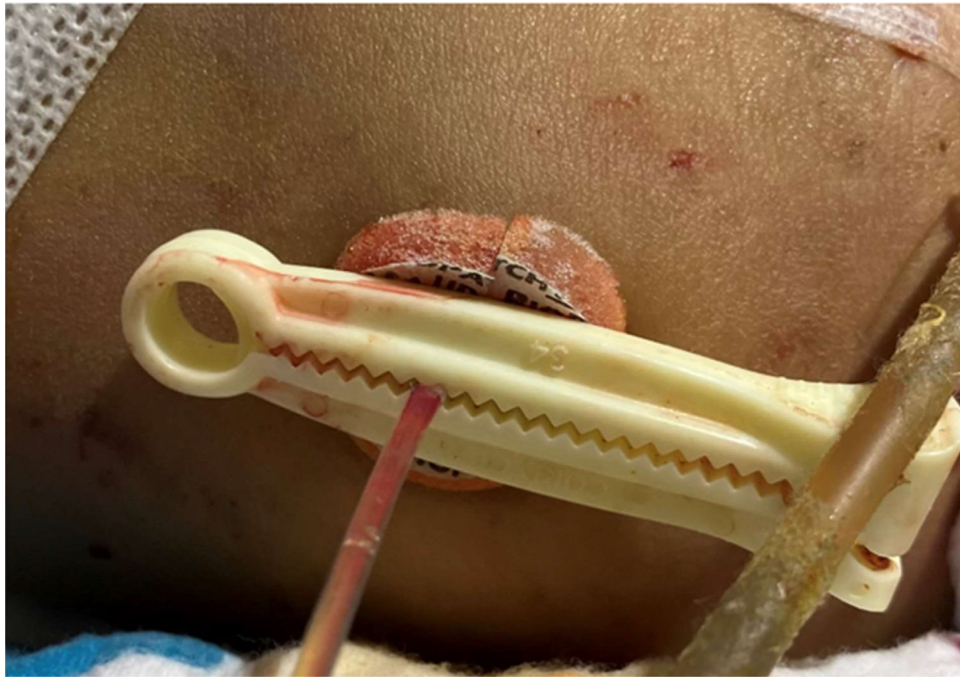
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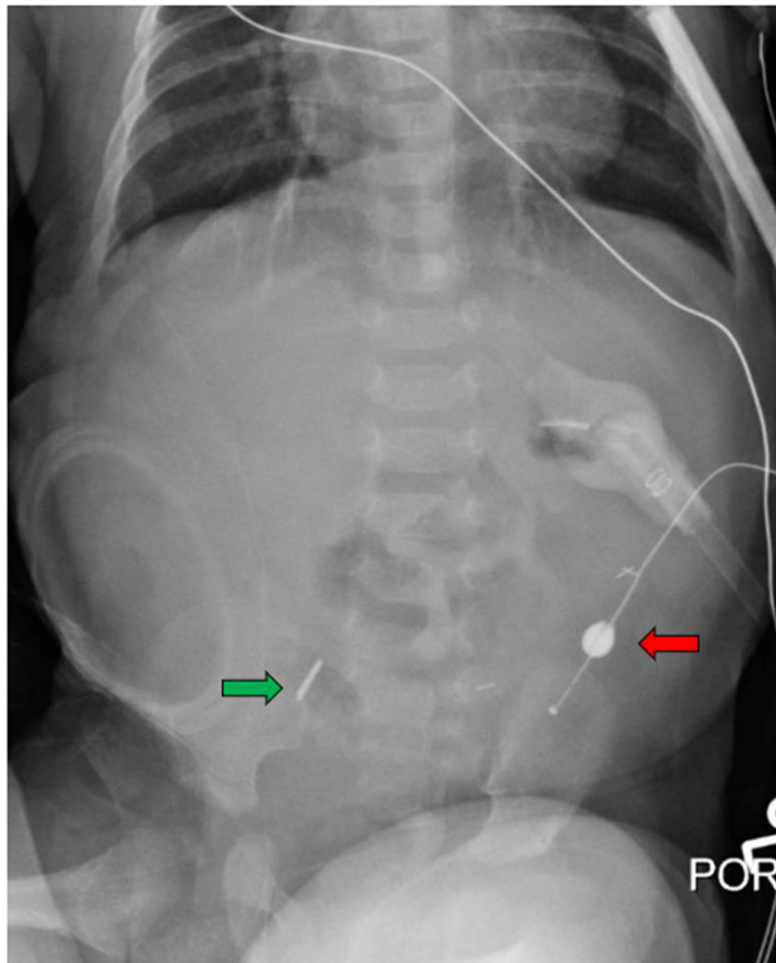
**Fig. 1. Intraoperative view of ileal tube placement.**

Ileal tube (red arrow) is placed in the most proximal end of the ileum with balloon inflated. Cecopexy (yellow arrow) performed, and clip placed on cecum (green arrow) for future reference.

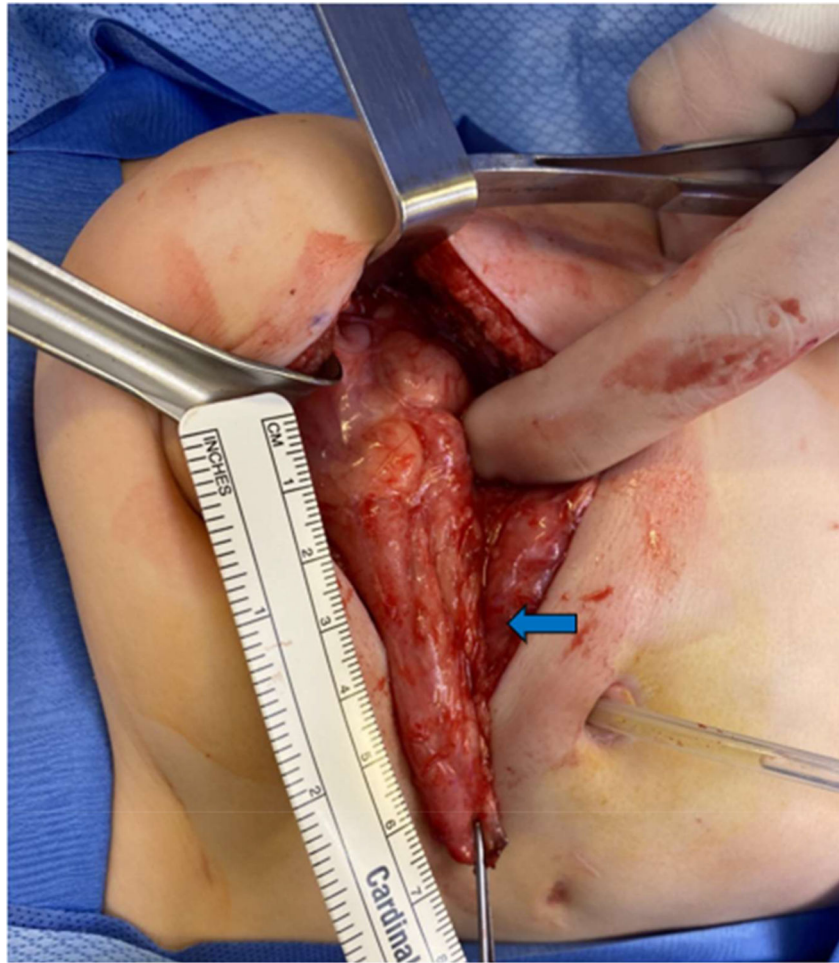


**Fig. 2. External ileal tube lengthening setup.**

The ileal tube is fixed externally on tension with an umbilical clamp. A standard Biopatch™ is placed between the abdominal wall and the umbilical clamp. Successive additional patches are added in a stacked fashion on the skin to distract the ileal tube until sufficient length is achieved.

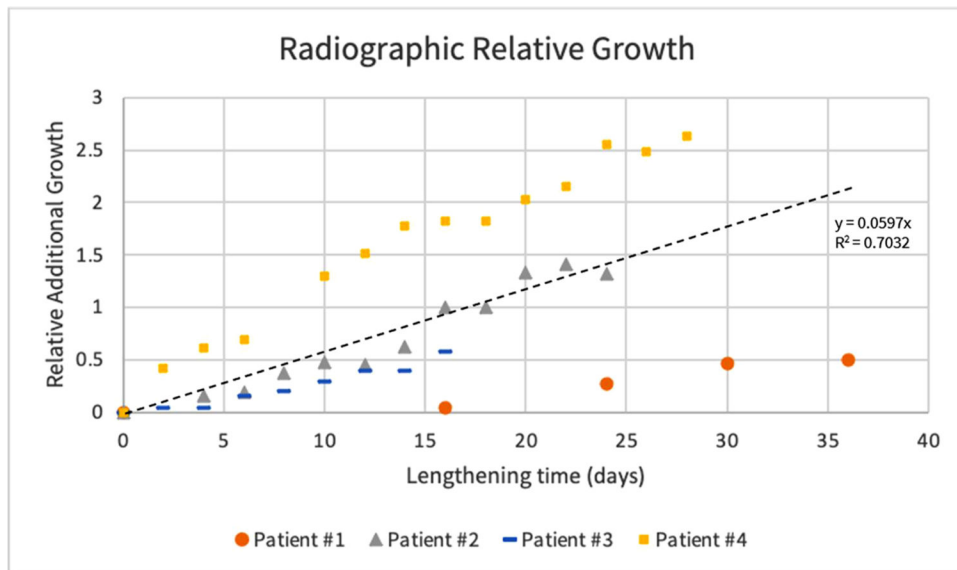


**Fig. 3. Postoperative radiograph after ileal tube lengthening.**  
Length of ileum measured between ileal tube (red arrow) and cecal clip (green arrow).



**Fig. 4. Ileal Length at restoration of continuity.**

After ileal lengthening is completed, enteric continuity is restored through ileo-enteric anastomosis. Final ileal length (blue arrow) is measured.



**Fig. 5. Radiographic Relative Additional Ileal Growth.**

Additional ileal growth from baseline as seen on radiographs plotted as a function of time. Measured distance between ileal balloon and cecal clip subtracted from baseline measurement to determine relative additional growth. Growth trajectory of each patient in the series plotted above. Linear regression best-fit line, equation, and coefficient of determination reported.

**Table 1**

**Patient Demographics.**

Age, diagnosis leading to short bowel syndrome, baseline ileal length, preoperative total intestinal length, and preoperative anatomic arrangement are presented for each patient in the series. NEC: necrotizing enterocolitis

Patient #	Age at Lengthening (days)	Diagnosis	Preoperative Ileal Length (cm)	Preoperative Total Intestinal Length (cm)	Intestinal Anatomy
1	339	Mesenteric mass	5	6	Total Discontinuity
2	71	NEC	1	24	End Jejunostomy
3	75	Atresia	6	36	End Jejunostomy
4	89	NEC	3	4	Total Discontinuity

**Patient Outcomes.**

Duration of active lengthening, baseline ileal length and additional length achieved, age at reanastomosis, ileal and total length at time of restoration of continuity with percentage gained, and complications for each patient reported. SSI: surgical site infection.

**Table 2**

Patient #	Duration of Lengthening (days)	Pre-lengthening Ileum (cm)	Radiographic Additional Length Achieved (cm)	Age at Restoration of Continuity (days)	Post-lengthening Ileum (cm)	Centimeters of Ileal Length Achieved (%)	Post-lengthening Total Intestinal Length (cm)	Complications
1	36	5	3.2	406	7	2 (40)	9	none
2	24	1	2.8	124	2	1 (100)	47	SSI
3	16	6	3.0	126	8	2 (33)	64	SSI
4	27	3	6.2	138	4.5	1.5 (50)	6.5	none