



Risk factors and outcomes of tapering surgery for small intestinal dilatation in pediatric short bowel syndrome



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ABSTRACT

Background: It remains unclear why in some short bowel syndrome (SBS) patients, the remaining small bowel (SB) dilates excessively leading to requirement of tapering surgery.

Methods: Among SBS children, we retrospectively analyzed risk factors for tapering surgery with logistic regression and compared the outcome of operated patients ($n = 16$) to those managed conservatively ($n = 44$) with Cox proportional hazards regression.

Results: SBS was caused by necrotizing enterocolitis (NEC) ($n = 31$), SB atresia (SBA) ($n = 13$), midgut volvulus ($n = 12$), or gastroschisis ($n = 4$). Patients with spontaneous symptomatic SB dilatation unable to wean parenteral nutrition (PN) underwent tapering surgery at median age of 1.04 (interquartile range 0.70–3.27) years. Missing ICV was related to an 8-fold ($p = 0.003$) increased risk while SBA diagnosis was related to a 13-fold risk of tapering surgery ($p < 0.001$). Increasing SB length and NEC diagnosis were protective of tapering ($p = 0.027$ – 0.004). Of operated patients, 75% reached enteral autonomy during follow-up and their postoperative adjusted PN weaning rate was similar to nonoperated children ($p = 0.842$).

Conclusion: SBS children with short remaining SB, missing ICV, and SBA etiology are more likely while NEC patients are less likely than others to necessitate tapering surgery. Postoperative PN weaning rates were comparable to patients who initially had more favorable intestinal anatomy and adapted without surgery.

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Intestinal failure (IF) in children is defined as reduction of functional intestinal mass below the minimum needed for sufficient absorption of fluid and nutrients, and requirement of long-term parenteral nutrition (PN) to sustain survival and maintain adequate growth instead [1,2]. The most common reason for IF among children is short bowel syndrome (SBS) caused by extensive small bowel (SB) resection, indicated during infancy for conditions such as necrotizing enterocolitis (NEC), midgut volvulus (MGV), SB atresia (SBA), and gastroschisis [3,4]. Increasing remaining bowel length, preserved ileocecal valve (ICV), and reestablishment of intestinal continuity improve the chances for weaning off PN, which is the main goal in the treatment of SBS patients [2,3,5,6].

After resection, the remaining SB undergoes structural and functional changes to improve its absorptive capacity [7,8]. In some patients, the remaining SB dilates as part of the physiological adaptation process

[3,7,8]. Although this increases the bowel absorptive surface, excessive SB dilatation may also impair intestinal motility and promote bacterial overgrowth (BO), which in turn predispose SBS patients to septic infections, D-lactate acidosis, and malabsorption [9,10]. We have recently demonstrated that adaptive SB dilatation is an independent risk factor for prolonged PN duration and decreased survival in SBS children [11]. In addition to PN dependence, anatomy of the remaining bowel and the etiology of SBS possibly influence the risk of excessive adaptive dilatation [10–13].

Tapering surgery of dilated SB in order to relieve the related functional problems has become increasingly popular during recent years especially in pediatric patients [14–20]. In serial transverse enteroplasty (STEP) and longitudinal intestinal lengthening and tapering (LILT), SB length is increased by narrowing the dilated intestine while preserving all absorptive mucosal surface [19,21]. Alternatively, dilated parts may be narrowed by simple tapering enteroplasty or segmental resection when the remaining bowel is not critically short [22]. SBS patients unable to wean from PN because of complications related to SB dilatation are considered as candidates for tapering surgery [15,17,19]. Among such patients, tapering increases the likelihood of weaning off PN possibly by reducing BO, by improving bowel motility, and by promoting absorption [14–20,23]. Tapering may also ameliorate liver dysfunction by decreasing the need for PN [24]. However, few studies have compared

Abbreviations: ALT, alanine aminotransferase; BO, bacterial overgrowth; HR, hazard ratio; ICV, ileocecal valve; IF, intestinal failure; IQR, interquartile range; LILT, longitudinal intestinal lengthening and tapering; MGV, midgut volvulus; NEC, necrotizing enterocolitis; OR, odds ratio; PN, parenteral nutrition; SB, small bowel; SBA, SB atresia; STEP, serial transverse enteroplasty; CI, confidence interval.

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the outcomes of SBS patients having undergone tapering to those treated without surgery [25], and it remains unclear how SBS children necessitating tapering differ from patients who adapt without surgical procedures.

Identification of risk factors for the need of tapering surgery would help to recognize the most susceptible SBS patients earlier and target particular attention at them. Moreover, as tapering surgery has become an established practice in the treatment of SBS, its efficacy should be evaluated not only by comparing the preoperative and postoperative results but also by paralleling the outcome of operated patients to those managed conservatively. To this end, we aimed to analyze risk factors for the need of tapering surgery for spontaneous adaptation-related excessive small intestinal dilatation, as well as to study the respective PN weaning and survival rates in SBS children treated with or without tapering surgery.

1. Methods

1.1. Patients

This was a retrospective study including all patients with IF caused by SBS treated by our intestinal rehabilitation program [26] in 2002–2015 ($n = 61$). Inclusion criteria were SB resection $>50\%$ of age-adjusted reference value, or duration of PN for over two consecutive months [4,27,28]. Patients with tapering surgery following controlled bowel expansion ($n = 1$) and other etiology of IF ($n = 33$), such as primary motility disorders, mucosal diseases, and lymphatic disorders were excluded. Medical records were reviewed for SBS etiology, gestational age and weight, duration of PN, plasma citrulline levels, intestinal anatomy, and SB width in contrast series. Bowel length was expressed both as absolute values and as percentage of age-adjusted reference values [28] while the presence of ICV and ileum was recorded as absent or present. Maximal width of SB perpendicular to the longitudinal axis of the bowel was measured in contrast SB series as described previously [11]. SB width was expressed both as mm (absolute SB width) and as internally standardized ratio to the height of the fifth lumbar vertebra (SB diameter ratio) [11]. At last follow-up, plasma alanine aminotransferase (ALT) and bilirubin levels as well as liver biopsy findings were recorded, and growth was assessed by using height z-scores as well as age-adjusted ISO-BMI for children aged 2–18 years; age-adjusted weight-to-height percentiles (based on national data) for children under 2 years of age; and the BMI for patients older than 18 years [26].

1.2. Tapering surgery

Tapering surgery was considered in SBS patients with excessive bowel dilatation who were unable to wean from parenteral support and presented with complications related to symptoms of SB dilatation, such as BO or dysmotility. The presence of one or several dilated SB segments clearly exceeding the diameter of normal-caliber SB in the same contrast SB series was a prerequisite for tapering surgery. BO and dysmotility were diagnosed based on clinical symptoms, including abdominal bloating, vomiting, increased intestinal secretions, and inability to advance enteral nutrition. Altogether 16 patients underwent tapering procedures, including STEP, LILT, tapering enteroplasty, and resection of dilated bowel segment. In this study, resection of dilated segment was included in tapering procedures because it was used as STEP, LILT, or tapering enteroplasty to treat excessively dilated SB, however, for patients with limited dilatation and sufficient remaining small intestinal length. Previously standardized surgical techniques for STEP and LILT were followed [17,19]. Simple tapering of a dilated bowel segment was performed with a linear stapler along the long axis of a dilated bowel segment [22]. Indications for tapering surgery and possible postoperative complications were recorded. Primary and retapering procedures were analyzed separately. One of the primary tapering procedures (STEP) had been performed in another hospital, after which the patient

was referred to us and later underwent a reoperation for symptomatic redilatation.

1.3. Statistical methods

Data are presented as medians with interquartile ranges (IQR) or as frequencies. Spearman rank correlation was used to examine associations between variables. Mann–Whitney U test was used to compare continuous variables, Wilcoxon signed-rank test to compare repeated measurements within groups, and Fisher exact test to compare frequencies between subgroups. We conducted simple and multiple logistic regression analyses to further estimate the magnitude of association between different variables and the need for tapering surgery. The odds ratios (ORs) with 95% confidence intervals (CIs) were computed, and statistically significant predictors from the simple regression model were chosen for the multiple regression analysis. As SB dilatation was a prerequisite for tapering surgery, SB width or SB diameter ratio were not included in these analyses. Kaplan–Meier plots with the log-rank test were used to compare the PN weaning rates of operated patients to nonoperated patients, and those receiving PN at the end of follow-up or who died while receiving PN were censored from the analysis. Finally, we studied the predictive values for different variables on cumulative rates of weaning off PN and survival by generating hazard ratios (HRs) with 95% CIs with Cox proportional hazards regression model. Variables showing statistically significant univariate associations with PN weaning rates were included in the multivariate model. The level of significance was set at $p < 0.05$ and all analyses were carried out with SPSS version 22.

1.4. Ethics

The study protocol was approved by the hospital ethical committee.

2. Results

2.1. Patient characteristics

We identified 60 eligible patients with IF caused by SBS (males 58%, $n = 35$). At the time of study, intestinal continuity had been reestablished in all and none had an enterostomy. Baseline diagnoses were necrotizing enterocolitis (NEC) ($n = 31$), midgut volvulus (MGV) ($n = 12$), small bowel atresia (SBA) ($n = 13$), and gastroschisis ($n = 4$) (Table 1). Four patients with both SBA and gastroschisis were analyzed in the SBA group. Median residual small bowel length was 40 (25–60) cm while colon length was 32 (24–41) cm, corresponding to 25% (17%–44%) and 93% (61%–100%) of age-adjusted reference values, respectively [28]. The ICV was missing in 28 (47%) and no ileum was remaining in 27 (45%). Median gestational age was 34 (28–36) weeks and birth weight was 2090 (960–3035) grams. Median plasma citrulline was 13 (10–17) before and 24 (20–29) after weaning off PN ($p < 0.001$).

2.2. Tapering surgery

Altogether 16 patients (27%) had undergone tapering procedures. In addition to bowel dysmotility, inability to increase enteral intake and BO were the most common indications for tapering surgery (Table 2). The patient having undergone bowel resection had been previously off PN before developing symptomatic segmental SB dilatation and becoming PN dependent again. Postoperative complications requiring relaparotomy occurred in two while milder complications in two patients (Table 2). Preoperative contrast SB series were performed on 10 patients while postoperative contrast SB series on 13 patients. The maximal SB diameter ratio in these contrast series decreased significantly following tapering (Table 2, $p = 0.009$), while no decrease in maximal absolute SB width was observed.

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