

Transanal endorectal pull-through procedure versus transabdominal surgery for Hirschsprung disease

A systematic review and meta-analysis

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Abstract

Objective: To analyze the clinical results of transanal endorectal pull-through (TERPT) and transabdominal approach (TAB) in the treatment of Hirschsprung disease.

Methods: We searched all publications in the PubMed, MEDLINE, EMBASE, and Cochrane library databases between January 2003 and November 2018. The study included randomized controlled trials (RCTs) and observational clinical studies (OCSs), to compare the surgery duration, length of postoperative hospital stay, incidence of postoperative incontinence/soiling, constipation, and enterocolitis between the TERPT and TAB groups. Mantel-Haenszel method was used for continuous variables, the combined odds ratios (ORs) and 95% confidence intervals (CIs) for dichotomous variables were used.

Results: In the 87 studies, we include 1 case of RCTs and 9 cases of OCSs. Including 392 cases of TERPT and 332 cases of TAB groups. TERPT has a short postoperative hospitalization [mean difference (MD) = -6.74 day; 95% Cls; -13.26 to -0.23; P = .04], and a low incidence of postoperative incontinence (ORs = 0.54; 95% Cls, 0.35-0.83; P = .006) and constipation (ORs = 0.50; 95% Cls, 0.28-0.90; P = .02). There was no difference in duration of surgery (MD = -30.59 min; 95% Cls, -98.01-36.83; P = .37) and incidence of postoperative enterocolitis (ORs = 0.78; 95% Cls, 0.53-1.17; P = .23).

Conclusion: TERPT is superior to TAB in terms of hospitalization time, postoperative incontinence, and constipation. However, there are still a large number of RCTs to verify, and more trials are expected to be testified in the future.

Abbreviations: CI = confidence interval, HD = Hirschsprung disease, MD = mean difference, NOS = Newcastle-Ottawa scale, OCS = observational clinical study, OR = odds ratio, RCT = randomized controlled trial, TAB = transabdominal approach, TERPT = transanal endorectal pull-through.

Keywords: constipation, enterocolitis, Hirschsprung disease, incontinence/soiling, transabdominal approach, transanal endorectal pull-through

1. Introduction

Hirschsprung disease (HD), a common congenital intestinal disease in children, is caused by continuous spasms of the intestinal tract due to a lack of ganglion cells in the colon and fecal stasis in the proximal colon, which corresponds to

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hypertrophy and expansion of the proximal colon. The common classification includes a short segment and long segment, and the main treatment is radical surgery in addition to some short segments. There are many surgical approaches to HD, including the transabdominal approach (TAB) and transanal endorectal pull-through (TERPT). The TAB includes 4 types: the Swenson, Duhamel, Rehbein, and Soave procedures.^[1,2]

The Swenson procedure involves the removal of the entire affected site and end-to-end anastomosis of the normal colonic anal canal. In the Duhamel procedure, the normal colon is pulled out through the posterior rectal space and anastomosed with the non-angular rectum on the contralateral side. The Rehbein procedure is a low rectal anastomosis in the pelvic cavity. In the Soave procedure, physiological saline is injected into the rectum after cutting through the rectal muscle layer in a circular manner, while keeping the mucosa intact to the dentate line level.

In 1998, De la Torre-Mondragon proposed a new treatment called single-stage TERPT,^[1] which is more suitable for infants. This minimally invasive surgery with an anal approach has become an increasingly popular method for the treatment of HD, eliminating the risk of complications such as abdominal adhesions and pelvic nerve injury. The advantages of TERPT include a good cosmetic effect and a short hospitalization time, and its safety has been proved by many studies.^[1,3] However, there are a variety of ways to choose surgery in clinical practice, and no consensus has been reached. In this study, a meta-analysis

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was conducted to better evaluate the effectiveness and reliability of TERPT and TAB and to provide a basis for future clinical diagnosis and treatment.

2. Materials and methods

2.1. Search strategy

Strictly abiding by the PRISMA guidelines, we searched all publications in the PubMed, MEDLINE, EMBASE, and Cochrane library databases between January 2003 and November 2018. We searched the terms "Hirschsprung's disease," "treatment of Hirschsprung's disease," "perineal approach," "abdominal approach," "fecal-constipation," and "pullthrough," and their combinations. These studies were evaluated for inclusion in our analysis after relevant titles and summaries were identified. We included all studies that met the inclusion criteria to compare TERPT and TAB results and searched the full text. A list of references for relevant studies was also searched to identify other possible studies that met the criteria. The search results included observational clinical studies (OCS) because of the lack of randomized controlled trials (RCTs) that addressed the study questions. Most studies measured functional outcomes by the incidence of postoperative complications. Some studies involved overlapping data, and we took steps to exclude the earlier data.

2.2. Inclusion and exclusion criteria

The inclusion criteria were as follows: patients were newborns, infants, or children; study participants were patients with HD undergoing surgical treatment; the type of study was an RCT or an OCS; TERPT was used in the observation group and TAB was used in the control group; the study compared one or more of the results of TERPT and TAB in the treatment of HD, including the duration of surgery, duration of postoperative hospitalization, and incidence of postoperative complications (postoperative incontinence/soiling, postoperative constipation, and postoperative enterocolitis); and only articles published in English.

The exclusion criteria were as follows: adult patients with HD; duplicate reporting; the data of the duration of surgery, duration of postoperative hospitalization, and the incidence of postoperative complications (postoperative incontinence/soiling, postoperative constipation, and postoperative enterocolitis) were all missing; and (4) article type was a letter, review, editorial, or case report.

2.3. Research selection and data extraction

The included studies were independently evaluated by 2 reviewers. When the 2 reviewers could not agree on study inclusion, a third reviewer was consulted. The following data were extracted from the literature: first author, year of publication, type of study, sample size, patient sex, interventional measures in the 2 groups, age at operation, follow-up time, and outcomes.

2.4. Outcome indicators

Observation and comparison of patients after TERPT and TAB included the following: duration of surgery; duration of postoperative hospitalization; incidence of postoperative incontinence/soiling; incidence of postoperative constipation; and incidence of postoperative enterocolitis. Our review was based on published research, and unpublished data were not available to the authors.

2.5. Quality evaluation

The OCSs were graded according to the Newcastle-Ottawa scale (NOS) evaluation criteria, including selection (4 points), comparability (2 points), and outcome/exposure (3 points). A full score was 9 points, \leq 4 points indicated low-quality literature, and \geq 5 points were considered high-quality literature.

The RCT was evaluated with the Jadad rating scale, including random sequence generation (2 points), randomized hiding (2 points), blinding (2 points), and loss of follow-up and withdrawal (1 point). The full score was 7, \leq 3 points indicated low-quality literature, and \geq 4 points were considered high-quality literature. The quality score was assessed by 2 independent reviewers. If they could not reach an agreement, the final score was determined by a third reviewer.

2.6. Statistical analysis

RevMan 5.3 (The Cochrane Collaboration, Oxford, United Kingdom) software was used for the meta-analysis. The Mantel-Haenszel method was used for continuous variables, and the combined odds ratios (ORs) and 95% confidence intervals (CI) were used for dichotomous variables. The difference was statistically significant when the *P* value was <.05. The pooled mean difference (MD) was measured in the meta-analysis using the inverse variance method. Inferred heterogeneity was determined according to I^2 . When I^2 was <50%, there was no obvious heterogeneity in the analysis, and the fixed effect model was used. When the I^2 was \geq 50%, there was significant heterogeneity among the analyses, and the random effect model was selected.

3. Results

3.1. Literature screening process

In 87 studies reporting on the results of the treatment of HD by the TERPT approach, 12 studies compared the results of treatment by the TERPT and TAB approaches and met the conditions for inclusion in our analysis. Two studies^[4,5] used different methods than the other studies to evaluate postoperative outcomes and were excluded. The remaining 10 studies were included, including 724 patients, 392 in the TERPT group and 332 in the TAB group (Fig. 1).

3.2. Quality evaluation of included studies

The scores of the OCSs ranged from 4 to 6, including $3^{[6-8]}$ of low quality and $6^{[9-14]}$ of high quality. This RCT^[15] was scored 3 for low-quality literature (Table 1).

3.3. Study characteristics

A total of 724 patients were included in the 10 studies; TERPT was performed in 392 cases and TAB in 332 cases (including 209 Soave cases in 5 studies, 96 Duhamel cases in 5 studies, and 21 Swenson cases in 1 study). Seven of the studies identified the sex of the patients, with >70% of patients being male. The age of operation was available for 8 studies. The follow-up duration was given in 6 studies. In addition to the research results required by our study, there were simple descriptions of other results in the 10 studies, including anastomotic leakage and pelvic abscess (Table 2).

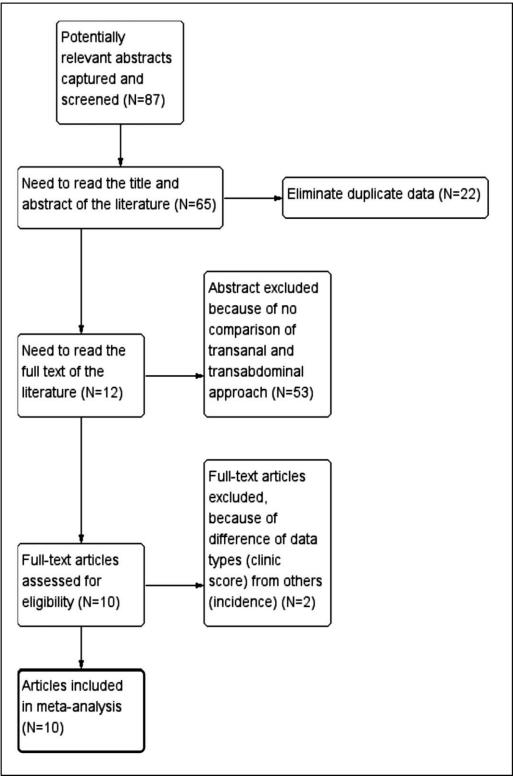


Figure 1. Flow chart showing the search strategy and search results.

3.4. Surgery duration

The studies of Sosnowska et al,^[10] Onishi et al,^[9] Tannuri et al,^[11] and Ishikawa et al^[12] reported the full data for the operative time (Table 3). There was no significant difference in the operative time

between the TERPT group and the TAB group (MD = -30.59 min, 95% CI -98.01-36.83, P=.37) (Fig. 2A), and the heterogeneity was significant ($I^2 = 91\%$) (Fig. 2A), so the random effect model was adopted. A funnel plot showed certain publication biases (Fig. 2B).

Quality ev	
Table 1	

Study (author, year)		Gunadi et al, 2018 ^[6]	Onishi et al, 2016 ^[9]	Sosnowska et al, 2016 ^[10]	Ishikawa et al, 2008 ^[12]
Selection (4 points)	Is the case definition adequate?	1	0	1	1
	Representativeness of the cases	1	1	1	1
	Selection of controls	0	0	1	0
	Definition of controls	0	1	0	0
Comparability (2 points)	Comparability of cases and controls on the basis of the design or analysis	1	1	1	1
Exposure (3 points)	Ascertainment of exposure	1	1	1	1
	Same method of ascertainment for cases and controls	0	1	1	1
	Nonresponse rate	0	0	0	0
Total score		4	5	6	5

(1 a)

Study (autho	r, year)	Tannuri et al, 2017 ^[7]	Tannuri et al, 2009 ^[11]	Martins et al, 2009 ^[10]	Kim et al, 2010 ^[3]	Stensrud et al, 2010 ^[14]
Selection (4 points)	Representativeness of the exposed cohort	0	1	1	1	1
,	Selection of the non exposed cohort	0	1	0	1	1
	Ascertainment of exposure	1	0	1	1	0
	Demonstration that outcome of interest was not present at start of study	0	0	0	0	0
Comparability (2 points)	Comparability of cohorts on the basis of the design or analysis	1	1	1	1	1
Outcome (3 points)	Assessment of outcome	1	1	1	1	0
	Was follow-up long enough for outcomes to occur	1	1	0	1	1
	Adequacy of follow-up of cohorts	0	1	0	0	1
Total score		4	6	4	6	5
(1.b)						
(1)						
Study (autho		sequence n (2 points)	Randomized hiding (2 points)	Blinding (2 points)	Loss of follow-up a withdrawal (1 poir	
Aslanabadi et	al, 2008 ^[15]	1	1	0	1	3

(2)

(1) Newcastle-Ottawa scale (2) Jadad rating scale (a) case-control study (b) cohort study.

3.5. Duration of postoperative hospitalization

The studies of Sosnowska et al,^[10] Onishi et al,^[9] Tannuri et al,^[11] and Aslanabadi et al^[15] reported the full data for the duration of postoperative hospitalization (Table 3). The TERPT group had shorter hospital stays than the TAB group (MD = -6.74 days, 95% CI -13.26 to -0.23, P = .04) (Fig. 3A), and the heterogeneity was significant ($I^2 = 94\%$) (Fig. 3A), so the random effect model was adopted. A funnel plot showed certain publication biases (Fig. 3B).

3.6. Postoperative incontinence/soiling

A total of 9 (1 RCT and 8 OCSs) studies^[7–15] reported postoperative incontinence/soiling (Table 3). Among them, 4 studies^[7,8,11,14] elaborated on the definition of incontinence/ soiling (Table 4). Overall analysis showed that the incidence of postoperative incontinence was lower in the TERPT group than for all transabdominal surgeries (OR=0.54, 95% CI 0.35–0.83, P=.006) (Fig. 4.1A) or for the Soave group (OR=0.52, 95% CI 0.29–0.92, P=.02) (Fig. 4.3A). However, when the TERPT and

Duhamel (OR=0.58, 95% CI 0.28–1.17, P=.13) (Fig. 4.2A) groups were compared, there was a slight difference between them. When TERPT and TAB were compared, the heterogeneity test showed no obvious heterogeneity (I^2 =0%) (Fig. 4.2A). Therefore, the fixed effect model was selected. A funnel plot showed certain publication biases (Figs. 4.1B, 4.2B, 4.3B).

3.7. Postoperative constipation

Postoperative constipation was reported in seven studies (Table 3).^[6,8,9,12–15] Two^[6,14] of these studies defined constipation (Table 4). The results showed that the incidence of constipation in the TERPT group was lower than that in the TAB group (OR=0.50, 95% CI 0.28–0.90, P=.02) (Fig. 5.1. A). There was no obvious heterogeneity in the analysis ($I^2 = 38\%$) (Fig. 5.1A). The incidence of postoperative constipation in the Soave group was higher than that in the TERPT group, but there was no statistically significant difference (OR=0.77, 95% CI 0.39–1.54, P=.46) (Fig. 5.2A), and the heterogeneity was low ($I^2 = 41\%$) (Fig. 5.2A). Therefore, the fixed effect

Table 2

Characteristics of studies included in the meta-analysis.

Study (author, year)	Study type	Setting	Group	Sex (No. of M) [n]%	Operation method	Age at operation, mean $\pm \text{SD}$ or mean or median	Follow-up time mean \pm SD or mean or median	Outcomes
Gunadi et al, 2018 ^[6]	Case-control study	SC	TERPT:21	31 (79.49)	Tansanal endorectal pull-through	NR	NR	Enterocolitis, constipation
			TAB:18		Soave, Duhamel, posterior neurectomy and posterior myectomy	NR	NR	
Onishi et al, 2016 ⁽⁹⁾	Case-control study	SC	TERPT:37	83 (78.30)	Tansanal endorectal pull-through	(159.00±252.06) D	11Y	Postoperative incontinence/soiling, enterocolitis, constipation, anastomotic leakage, leakage from biopsy site, stenosis of muscular cuff, achalasia, wound infection, mucosal prolapse, ileus, bleeding of muscular cuff
			TAB:69		SD open approach, one-step modified Soave procedure)	(404.66±826.09) D	6.1Y	
Sosnowska et al, 2016 ^[10]	Case-control study	SC	TERPT:10	24 (82.76)	Tansanal endorectal pull-through	16M	NR	Incontinence/soiling, enterocolitis, obstruction, stoma prolapse
			TAB:19		Duhamel-Martin	49M	NR	
Tannuri et al, 2017 ^[7]	Cohort study	SC	TERPT:21	30 (73.17)	Tansanal endorectal pull-through	10 (10D-72M)M	26 (6–55) M	Incontinence/soiling
			TAB:20		Duhamel	41 (6M-110M)M	30 (6-60) M	
Tannuri et al, 2009 ^[11]	Cohort study	SC	TERPT:35	NR	Tansanal endorectal pull-through	(11.0±15.1) M	(28.4±20.6) M	Incontinence/soiling, enterocolitis, wound infection
			TAB:29		Duhamel	(42.0 ± 34.8) M	(60.5±44.4) M	
Aslanabadi et al, 2008 ^[15]	RCT	SC	TERPT:21	35 (83.33)	Tansanal endorectal pull-through	(14.2±35.4) M	At least 12 mo	Incontinence/solling, enterocolitis, constipation, intraoperative bleeding (necessitating blood transfusion), second laparotomy needed, leak from anastomosis, pelvic abscess, recurrent constipation
			TAB:21		Swenson	(16.4±33.5) M		
Ishikawa et al, 2008 ^[12]	Case-control study	SC	TERPT:8	NR	Tansanal endorectal pull-through	NR	NR	Enterocolitis, constipation, incontinence/soiling
			TAB:20		Soave	NR	NR	
Martins et al, 2009 ^[8]	Cohort study	SC	TERPT:19	36 (85.71)	Tansanal endorectal pull-through	60M	NR	Constipation, incontinence/soiling
(0)			TAB:23		Duhamel	104,4M	NR	
Kim et al, 2010 ^[3]	Cohort study	MC	TERPT:192	234 (83.27)	Tansanal endorectal pull-through	(5.8±1.1) M	(67.9±2.2) M	Enterocolitis, constipation, incontinence/soiling
			TAB:89		Soave	(13.5±2.3) M	(101.2±4.8) M	
Stensrud et al, 2010 ^[14]	Cohort study	SC	TERPT:28	NR	Tansanal endorectal pull-through	2.2M	57 (15–92) M	Enterocolitis, constipation, incontinence/soiling
			TAB:24		Soave	13M	109 (81–126) M	

D=day, M=month, MC=multicenter, NR=not report, RCT=randomized controlled trial, SC=single center, TAB=transabdominal approach, TERPT=transanal endorectal pull-through.

model was selected. A funnel plot showed certain publication biases (Figs. 5.1B, 5.2B).

3.8. Postoperative enterocolitis

Eight studies^[6,9–15] reported postoperative enterocolitis, but only 1^[10] reported the total incidence of postoperative enteritis in the 2 groups, so this study was not included in the postoperative enterocolitis analysis (Table 3). Two studies^[6,11] defined enterocolitis (Table 4). The results showed that the incidence of postoperative enterocolitis in the TERPT group tended to be less than that in the TAB group, but there was no statistically significant difference (OR=0.78, 95% CI 0.53-1.17, P=.23) (Fig. 6.1.A). There was no obvious heterogeneity in the analysis $(I^2 = 47\%)$ (Fig. 6.1A). The incidence of postoperative enterocolitis in the Soave group was higher than that in the TERPT group, but the difference was not statistically significant (OR = 0.73, 95% CI 0.47–1.13, P=.16) (Fig. 6.2A), and the heterogeneity was low $(I^2 = 42\%)$ (Fig. 6.2A). Therefore, the fixed effect model was selected. A funnel plot showed certain publication biases (Figs. 6.1B, 6.2B).

4. Discussion

We analyzed 10 studies, including 1 RCT and 9 OCSs. The Jadad rating scale was used for the RCT, and the NOS rating scale was used for the OCSs. Six studies were found to be of high quality and 4 studies, including the RCT, were of low quality. Owing to the limitations of the research level, RCTs are less common, and the blinding method is difficult to implement. OCSs are more commonly performed, which affects the score and quality of the paper. All these shortcomings undermine the level of evidence in existing studies.

In the study of Chen et al,^[1] compared to the TAB group, the TERPT group had a shorter hospital stay and less postoperative incontinence/soiling and constipation. They found that the incidence of postoperative enterocolitis was similar after the 2 methods, unlike our results. In our study, there was no significant difference in operative time between the TERPT group and the TAB group; however, in the study of Chen et al, the TERPT group had a short operative time. In addition, in their meta-analysis, part of the study compared the TERPT and Soave methods. Our study not only compared the TERPT and

Table 3

Study (author, year)	Group	Operative time, min (mean \pm SD)	Postoperative hospital stay, day (mean \pm SD)	Incontinence/soiling N (%)	Constipation N (%)	Enterocolitis N (%)
Gunadi et al, 2018 ^[6]	TERPT:21	NR	NR	NR	1 (4.76)	3 (14.29)
	TAB:18	NR	NR	NR	5 (27.78)	3 (16.67)
Onishi et al, 2016 ^[9]	TERPT:37	265.92±108.42	17.78±7.23	0 (0.00)	0 (0.00)	4 (10.81)
	TAB:69	225.79±106.95	24.04 ± 8.28	0 (0.00)	0 (0.00)	12 (17.39)
Sosnowska et al, 2016 ^[10]	TERPT:10	230 ± 62.5	17 ± 1.75	0 (0.00)	NR	2 (6.90)
	TAB:19	240 ± 60	17±12.75	0 (0.00)	NR	
Tannuri et al, 2017 ^[7]	TERPT:21	NR	NR	17 (80.95)	NR	NR
	TAB:20	NR	NR	15 (75.00)	NR	NR
Tannuri et al, 2009 ^[11]	TERPT:35	120 ± 29.2	4.3 ± 3.69	7 (20.00)	NR	7 (20.00)
	TAB:29	232 ± 82.7	8.4 ± 6.30	12 (41.38)	NR	1 (3.45)
Aslanabadi et al, 2008 ^[15]	TERPT:21	NR	3.05 ± 0.86	1 (4.76)	0 (0.00)	1 (4.76)
	TAB:21	NR	18.67 ± 5.93	2 (9.52)	1 (4.76)	5 (23.81)
Ishikawa et al, 2008 ^[12]	TERPT:8	258 ± 48	NR	0 (0.00)	0 (0.00)	1 (12.50)
	TAB:20	294 <u>+</u> 48	NR	9 (45.00)	0 (0.00)	5 (25.00)
Martins et al, 2009 ^[8]	TERPT:19	NR	NR	5 (26.32)	1 (5.26)	NR
	TAB:23	NR	NR	9 (39.13)	8 (34.78)	NR
Kim et al, 2010 ^[3]	TERPT:192	NR	NR	20 (10.42)	16 (8.33)	65 (3.125)
	TAB:89	NR	NR	16 (17.98)	12 (13.48)	40 (44.94)
Stensrud et al, 2010 ^[14]	TERPT:28	NR	NR	15 (53.57)	7 (25.00)	7 (25.00)
	TAB:24	NR	NR	14 (58.33)	4 (16.67)	1 (4.17)

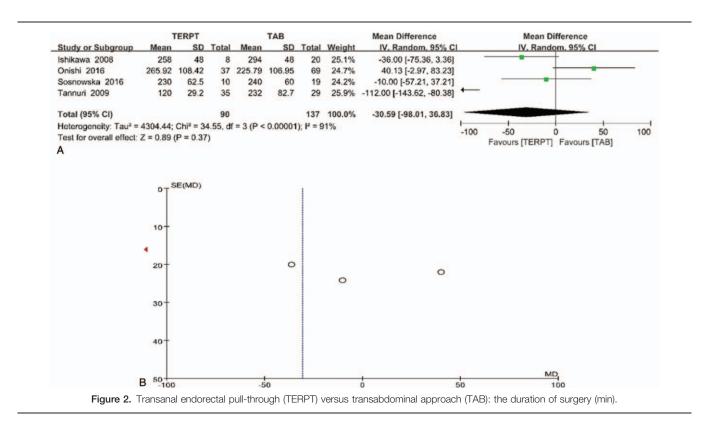
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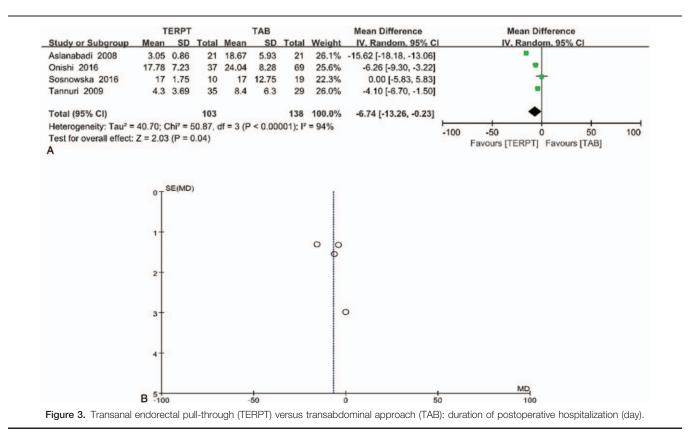
NR = not report, TAB = transabdominal approach, TERPT = transanal endorectal pull-through.

Soave methods, but also compared the TERPT and Duhamel methods; thus, our study was more comprehensive. Chen et $al^{[1]}$ observed a large amount of heterogeneity, which was similar to our study.

TERPT surgery is a less invasive surgical method with a good cosmetic effect and avoids interference in the abdominal cavity.^[16] Most studies indicated that the postoperative hospi-

talization time^[9–11,15] in the TERPT group was shorter than that in the TAB group; the heterogeneity test indicated that there was significant heterogeneity among the studies. However, there was no significant difference in the operative time between the 2 groups. In view of the heterogeneity of the operation time, we considered that it might be related to the various types of TAB surgery as well as the existence of a colostomy in preoperative





patients, the operation timing, the judgment and selection of surgical indications, the degree of difficulty of the operation, and the experience of the operator. The heterogeneity of the postoperative hospital stay was related to the differences in physician treatment plans, postoperative care, and discharge standards in different medical institutions.

The most common postoperative complications of HD are incontinence/soiling, constipation, and enterocolitis. In this study, we analyzed several types of open procedures (Swenson, Soave, Duhamel) in the TAB group. Four studies^[9,12–14] compared the incidence of postoperative incontinence/soiling, postoperative constipation, and postoperative enterocolitis after the TERPT and Soave surgeries. Four studies^[7,8,10,11] compared the incidence of urinary incontinence/contamination after the TERPT and Duhamel surgeries.

During anal traction, the anal sphincter may be overstretched, leading to incontinence/soiling. In this study, the incidence of postoperative incontinence/soiling in the TERPT group was lower than that in the TAB group; however, the detection of postoperative incontinence showed minimal heterogeneity $(I^2 =$ 0), suggesting that the lack of a consistent definition made little difference to our findings. It was found that although Soave was similar to TERPT, the TERPT operation could still significantly reduce incontinence/soiling compared to Soave. Related studies have shown that postoperative sphincter function does not decrease in patients undergoing TERPT.^[8,17] It may be that the risk of autonomic nerve damage is greatly reduced by avoiding pelvic dissection during TERPT operations. The occurrence of incontinence/soiling after the Duhamel operation is similar to that after the TERPT operation, which may be attributed to accumulated experience, where clinical surgeons realize the problem of traction strength and adjust the operation to minimize anal traction and extension.

Compared to TAB, the incidence of postoperative constipation was lower in the TERPT group. When comparing postoperative constipation, we found moderate heterogeneity ($I^2 = 38\%$), which may be attributed to different diagnostic criteria for enterocolitis in different centers. There was no significant difference in the incidence of constipation between the Soave group and the TERPT group. In the included studies, no relevant data were reported in the studies related to Duhamel surgery, but it was suggested in the relevant reports that the incidence of constipation after Duhamel surgery was relatively high.^[18] Surgeons took different measures to decrease the incidence of constipation.^[19]

The reported probability of postoperative enterocolitis was 4.6% to 54%. Studies have shown that the incidence of both postoperative enterocolitis and recurrent enterocolitis was significantly reduced.^[20] Our study also showed no obvious difference in the incidence of postoperative enteritis between the TERPT group and the TAB group or between the TERPT group and the Soave group. When comparing postoperative enterocolitis, we found that the heterogeneity was moderate (I^2 =47%), which may be attributed to the different diagnostic criteria established for enterocolitis in different centers.

5. Limitations

This study has many limitations, as follows. Limited language bias. The literature of other languages was not included in this study, which may lead to the omission of relevant studies in other languages. Publication bias. Firstly, only 10 articles were included in this meta-analysis. Furthermore, <10 studies were

Table 4

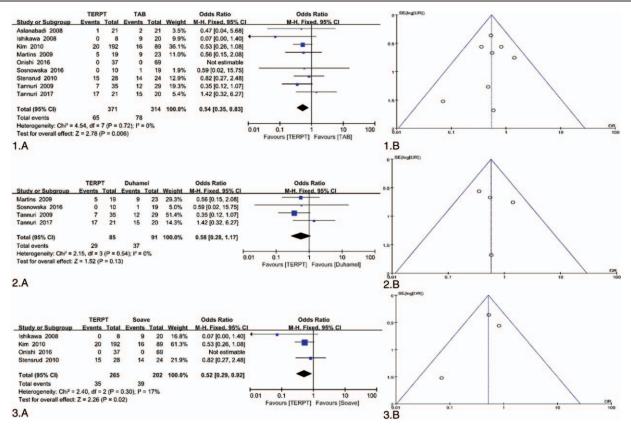
Study (author, year)	Definition of Incontinence/soiling	Definition of constipation	Definition of enterocolitis
Gunadi et al, 2018 ^[6]	NR	Constipation was defined based on Krickenbeck classification. Grade 1: manageable with diet; grade 2: requires laxatives; grade 3: resistant to diet and laxative	The enterocolitis diagnosis was determined using the Delphi score system
Onishi et al, 2016 ^[9]	NR	NR	NR
Sosnowska et al, 2016 ^[10] Tannuri et al, 2017 ^[7]	NR The FCI was based on the Clinical Evaluation of Fecal Continence (Holschneider criteria). The FCI questionnaire consists of 8 questions of easy comprehension regarding daily activities (questions 1–5); it also contains questions about diarrhea, constipation, and the use of supportive treatments (questions 6–8) according to the Holschneider criteria. The final score ranges from 0 to 16, with a value of 0 to 2 points for each question. The 0 to 5 range indicates poor continence, 6 to 10 indicates fair continence, and the maximum score of 16 indicates normal fecal	NR NR	NR
Tannuri et al, 2009 ^[11]	continence In patients older than 3 years, complete continence was defined as spontaneously evacuated soft stools, and there were no diurnal or nocturnal fecal soiling. Partially continent was defined as voluntary evacuations and few episodes of fecal soiling.	NR	Enterocolitis episodes were defined abdominal distension with loose offensive stool and general malais that had been treated by rectal washout and intravenous gentami and metronidazole.
Aslanabadi et al, 2008 ^[15]	NR	NR	NR
Ishikawa et al, 2008 ^[12]	NR	NR	NR
Martins et al, 2009 ^[8]	Martin's continence criteria based on the number of defecations, stool consistency, anal inspection and digital rectal examination	NR	NR
Kim et al, 2010 ^[3]	NR	NR	NR
Stensrud et al, 2010 ^[14]	In patients aged >3 years, soiling was defined as involuntary leaking of small amounts of stool, requiring change of underwear or diapers. Overflow incontinence was excluded by treating with laxative.	Constipation was defined based on Krickenbeck classification. Grade 1: manageable with diet; grade 2: requires laxatives; grade 3: resistant to diet and laxative	NR

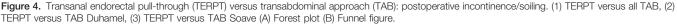
FCI=fecal continence index, NR=not report.

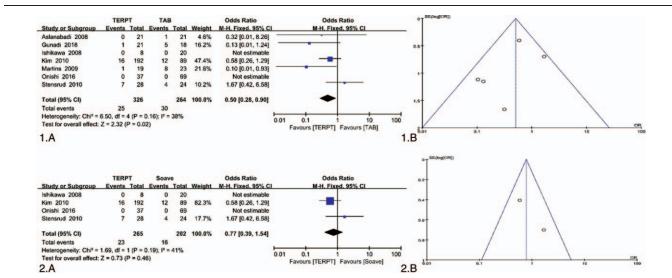
included in each result analysis. Secondly, owing to the lack of a large number of rigorous RCTs, most of the included studies were OCSs, which may have exaggerated the effect. There were other confounding factors, such as differences in the surgical indications and standards of each study, differences in open surgical methods, and the differences in discharge standards. Choosing fixed outcomes may be considered a limitation of the study, and any prediction of other outcomes (such as the incidence of anastomotic leakage or pelvic abscess) is inevitably misleading because it is impossible to determine all studies reporting on these outcomes. Reliability is also affected by differences in the definition of findings. In these studies, the definitions of postoperative incontinence, postoperative constipation, and postoperative enterocolitis were not standardized (Table 3).

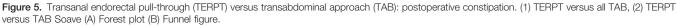
6. Conclusion

In summary, TERPT has a short postoperative hospitalization time and a low incidence of postoperative incontinence/soiling and constipation in the treatment of pediatric HD. However, the lack of a large number of RCTs in our included studies may have affected our results. Secondly, further studies are needed to repeatedly prove and evaluate the optimal surgical method to provide a basis for choosing the surgical method with the least impact on patients and the best effect.









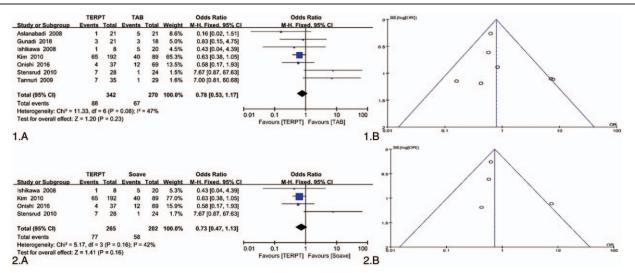


Figure 6. Transanal endorectal pull-through (TERPT) versus transabdominal approach (TAB): postoperative enterocolitis. (1) TERPT versus all TAB, (2) TERPT versus TAB Soave (A) Forest plot (B) Funnel figure.

Author contributions

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