A comparison of one-stage transanal endorectal pull-through and Duhamel pull-through operations for Hirschsprung's disease

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A comparison of one-stage transanal endorectal pull-through and Duhamel pull-through operations for Hirschsprung's disease

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Finally, I express my gratitude to my wife, who always believes and loves me. I can not spend time together because I am busy, I am deeply sorry and I love you. My nine month old son, Hyun, who can not stand alone yet, give my love and gratitude. I am a deficient father, but I will try to be *'a good father'* who is not ashamed.

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Chi Hwan Cha

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ABSTRACT

A comparison of one-stage transanal endorectal pull-through and Duhamel pull-through operations for Hirschsprung's disease

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Purpose: This study aimed to compare the operative results of the one-stage transanal endorectal pull-through operation (TERPT) with those of the Duhamel pull-through operation (DPT) for Hirschsprung's disease, including long-term functional outcomes.

Methods: Clinical data and postoperative courses of Hirschsprung's disease patients who had aganglionic bowel confined to the rectosigmoid and who underwent TERPT or DPT prior to 1 year of age between 2001 and 2013 at Severance Children's Hospital were reviewed and analyzed.

Results: Fifty-one patients underwent TERPT, and 50 patients underwent DPT. Age at the time of the pull-through operation is significantly younger in the TERPT group (1.7 ± 1.9 vs. 4.0 ± 2.4 months, $p<0.001$), and the mean operation time of TERPT was significantly shorter than that of DPT (154.6 ± 52.4 vs. 196.6 ± 65.0 min, $p=0.001$). Operation-related complications among those in the TERPT group were significantly fewer than among those in the DPT group (0 vs. 18%, $p=0.001$). However, hospital stays following the operation did not significantly differ (9.6 ± 3.1 vs. 11.4 ± 9.3 days, $p=0.200$). The readmission rate was also significantly lower in the TERPT group (39.2 vs. 64.0%, $p=0.013$). With respect to the long-term functional outcome, the

TERPT group had a significantly lower incidence of soiling (4.3 vs. 43.2%, $p<0.001$) and constipation (2.1 vs. 16.2%, $p=0.040$).

Conclusion: This study results showed significantly better postoperative clinical outcomes in the TERPT group. These results support the superiority of the TERPT procedure over DPT.

Key words: Hirschsprung's disease, transanal endorectal pull-through, Duhamel operation

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I. INTRODUCTION

Hirschsprung's disease is caused by the absence of ganglion cells in the submucosal plexus and myenteric plexus during the development of the enteric nervous system, which is characterized by intestinal obstruction in the aganglionic segment. In 1886, a Danish pediatrician Harald Hirschsprung reported two cases constipation in newborns due to dilatation and hypertrophy of the colon and this disease was named after his name.[1,2] At that time, most of the patients who had undergone extensive proximal colectomy had died of malnutrition or enterocolitis due to insufficient pathologic understanding of the disease. In 1901, Tittel first reported that ganglion cells were not observed in the distal colon of a patient with Hirschsprung's disease.[3] In 1946, Ehrenreis concluded, from a study of the clinical and roentgenologic onset and early development of the disease in 10 newborn infants, that the typical megacolon was not congenital but developed secondarily to the constipation.[4] No gross morphologic mechanical cause of constipation was found; it was defined as a primary dysfunction presumably of neurogenic origin. About half a year later, Whitehouse and Kernohan demonstrated that absence of ganglion cells of the myenteric plexus in the narrow distal segment was the underlying pathology.[5]

In the surgical treatments of Hirschsprung's disease, transabdominal pull-through operations have been the standard procedures since the first successful operation was introduced by Swenson in 1948.[6] Following the Swenson procedure, Duhamel and Soave also reported their methods of transabdominal pull-through, and all these methods evolved, with minor modifications.[7,8] Each of those three transabdominal pull-through operations has characteristic advantages over other operations, so the choice of operation methods has been a matter of pediatric surgeons' preferences.[9-11]

However, in the late of 1990s, the introduction of the one-stage transanal endorectal pull-through operation (TERPT) changed ideas, and this became the favorite operation method of pediatric surgeons for addressing Hirschsprung's disease.[12,13] The advantages of TERPT are well known, namely, that it does not require an abdominal incision, and there should be no surgery scar. It is a minimally invasive procedure that could minimize intraabdominal dissection and the risk of damage to pelvic structures.

Nevertheless, the better functional outcomes of TERPT procedures compared to those of the traditional abdominal pull-through operation have been established yet and controversial. A few studies have reported the superiority of TERPT over transabdominal pull-through operations, but in long-term follow-ups, no statistical significance has been found in comparisons of the functional outcomes.[14-16]

In my institute, Duhamel pull-through operation (DPT) was traditionally used, but in 2003, TERPT was introduced, and now both methods have been used. These circumstances lent themselves to a comparison of TERPT and DPT for ideal surgical treatment method of Hirschsprung's disease. Therefore, the aim of this study was to compare the operative results of TERPT and DPT, including long-term functional outcomes.

II. MATERIALS AND METHODS

The clinical data and postoperative courses of 108 Hirschsprung's disease patients who had aganglionic bowel confined to rectosigmoid and underwent TERPT (56) or DPT (52) at Severance Children's Hospital before the age of 1 year, between 2001 and 2013 were reviewed. Of these patients, 5 cases of Down syndrome (3 in TERPT, 2 in DPT) and 2 cases of technical failure in TERPT were excluded from the study. These two cases occurred at the beginning period of the TERPT procedure when was technically inexperienced. In DPT, 9 cases of the one-stage operation without colostomy were included in the study. Finally, the data of 101 Hirschsprung's disease patients, 51 cases from TERPT and 50 cases from DPT were reviewed. In the TERPT group, 31 cases were operated before 2010, and 20 cases were operated after 2010. In the DPT group, 40 cases were operated before 2010, and 10 cases were operated after 2010. All operations were performed by fully eligible 5 pediatric surgeons of Severance Children's hospital.

This study was approved by the Institutional Review Board of Severance hospital (4-2017-0435). This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors. There are no conflicts of interest to declare.

1. Surgical techniques

The surgical technique of TERPT was based on descriptions in previous literatures.[12,13] The patient was placed in the supine lithotomy position. Anal retraction was performed using a colostomy ring and silk sutures. The mucosa was incised 0.5 to 1 cm above the dentate line, and a circumferential submucosal dissection was carried out proximally. Excision of the muscle cuff to make a short muscle cuff was not performed. The rectal cuff was divided by a V-shape on the posterior rectal wall. When the transition zone was reached, a cryostat section evaluation of the biopsy was conducted to confirm the presence

of ganglion cell. Then, the bowel was transected, and anastomosis was performed.

In DPT, the classical surgical technique of DPT was adopted, which was described in the textbook.[17] All patients underwent laparotomy for DPT. Either a classical GIA stapler or laparoscopic GIA stapler for the retrorectal pull-through anastomosis, was used depending on the surgeons' preferences.

2. Assessment of postoperative functional outcome

The postoperative functional outcomes by checking for the presence of constipation or soiling in the patients who were older than 3 years at the time of the last follow-up was evaluated using the Krickenbeck classification[18] for classifying the grade of constipation and soiling (Table 1). Grades of constipation consist of grade 1 (manageable with diet), grade 2 (requires laxatives), and grade 3 (resistant to diet and laxatives). Grades of soiling also consist of grade 1 (occasionally), grade 2 (every day, no social problem), and grade 3 (constant, social problem).[19] All patients had normal voluntary bowel movements, so it was not included in the assessment of postoperative functional outcome.

Table 1. Krickenbeck classification

1. Voluntary bowel movements Feeling of urge, capacity to verbalize, hold the bowel movement	Yes/no
2. Soiling Grade 1: occasionally (once or twice per week) Grade 2: every day, no social problem Grade 3: constant, social problem	Yes/no
3. Constipation Grade 1: manageable with diet Grade 2: requires laxatives Grade 3: resistant to diet and laxatives	Yes/no

3. Statistical analysis

All data were analyzed using the statistical software IBM SPSS version 23 (IBM Co., Armonk, NY, USA). Two-sample t-tests and Chi-square tests were used to analyze the data, and p-values<0.05 were considered statistically significant.

III. RESULTS

1. Demographics

The characteristics of patients were summarized in Table 2. There were no statistically significant differences between the TERPT and DPT groups in sex ratio, birth weight, or gestational age. However, age at the time of the pull-through operation is significantly younger in the TERPT group. Consequently, the body weight at the time of the operation was significantly less in the TERPT group.

Table 2. Characteristics of patients

Characteristics	TERPT	DPT	P
Total number of cases	51	50	
Sex (M:F)	4.1:1 (41:10)	2.8:1 (37:13)	0.444
Birth weight (kg)	3.3 ± 0.5	3.2 ± 0.5	0.503
Gestational age (weeks)	38.7 ± 1.7	39.1 ± 1.4	0.226
Age at operation (months)	1.7 ± 1.9	4.0 ± 2.4	<0.001
Weight at operation (kg)	4.2 ± 1.4	6.4 ± 1.6	<0.001

2. Operative outcomes

The operative outcomes were summarized in Table 3. The mean operation time of TERPT was significantly shorter than that of DPT. Operation-related complications did not occur in the TERPT group, but the DPT group had more complications. In the DPT group, there was a case of postoperative death. The cause of death was ischemic brain injury due to cardiac arrest on the day of surgery.

The mean postoperative hospital stay was 9.6 days in the TERPT group and 11.4 days in the DPT group, but this difference was not found to be statistically significant.

Table 3. Operative outcomes

	TERPT (n=51)	DPT (n=50)	P
Operation time (min)	154.6 ± 52.4	196.6 ± 65.0	0.001
Operation-related complications	0	9 (18%)	0.001
Wound infection		6	
Intestinal obstruction		1	
Anastomosis leakage		1	
Death		1	
Postoperative hospital stay (day)	9.6 ± 3.1	11.4 ± 9.3	0.200

3. Postoperative outcomes

The postoperative outcomes were summarized in Table 4 and Figure 1.

A. Readmission after operation

In the TERPT group, 20 patients were readmitted for a total of 37 times. The majority of patients (14 patients) were readmitted once, and the most common cause of readmission was enterocolitis. During the postoperative 6 months, 80% (16 cases) of patients were readmitted and 73.0% (27 times) of total readmissions happened during the postoperative 6 months (Fig.1).

In contrast, readmission in the DPT group was significantly higher than in the TERPT group. In the DPT group, 32 patients were readmitted for a total of 85 times. Most patients (68.8%) were readmitted more than once. Enterocolitis was also the most common cause of readmission, but constipation as a cause of readmission was more common than in the TERPT group. Re-admission was more common after one year after surgery. Nearly two thirds (62.5%, 20 cases) of the patients were re-admitted after 1 year, and near one half (50.6%, 43 times) of total readmissions happened more than 1 year after the operation (Fig.1).

In the TERPT group, readmission due to enterocolitis occurred 36 times and readmission due to constipation occurred once. In the DPT group, there were 62 readmissions due to enterocolitis, 16 readmissions due to constipation, 1 readmission due to intestinal obstruction, 6 readmissions due to other causes (fusion of divided septum, fecal incontinence, rectal fistula, anal achalasia and abscess formation).

Table 4. Follow-up results

	TERPT (n=51)	DPT (n=50)	P
Postoperative follow-up period (month)	77.8 ± 37.9	78.7 ± 53.9	0.923
Readmission			
Patient (%)	20 (39.2)	32 (64.0)	0.013
Number of readmission	37	85	
1	14	10	
2	2	10	
3	3	3	
4≤	1	9	
Cause of readmission			
Enterocolitis	36	62	
Constipation	1	16	
Intestinal obstruction		1	
Etc.		6	
Reoperation			
Patient (n)	3	23	<0.001
Total occurrence	6	44	
Myectomy	4	14	
Re-do pull-through	1	4	
BOTOX® injection	1	12	
Septotomy	0	6	
Etc.		8	

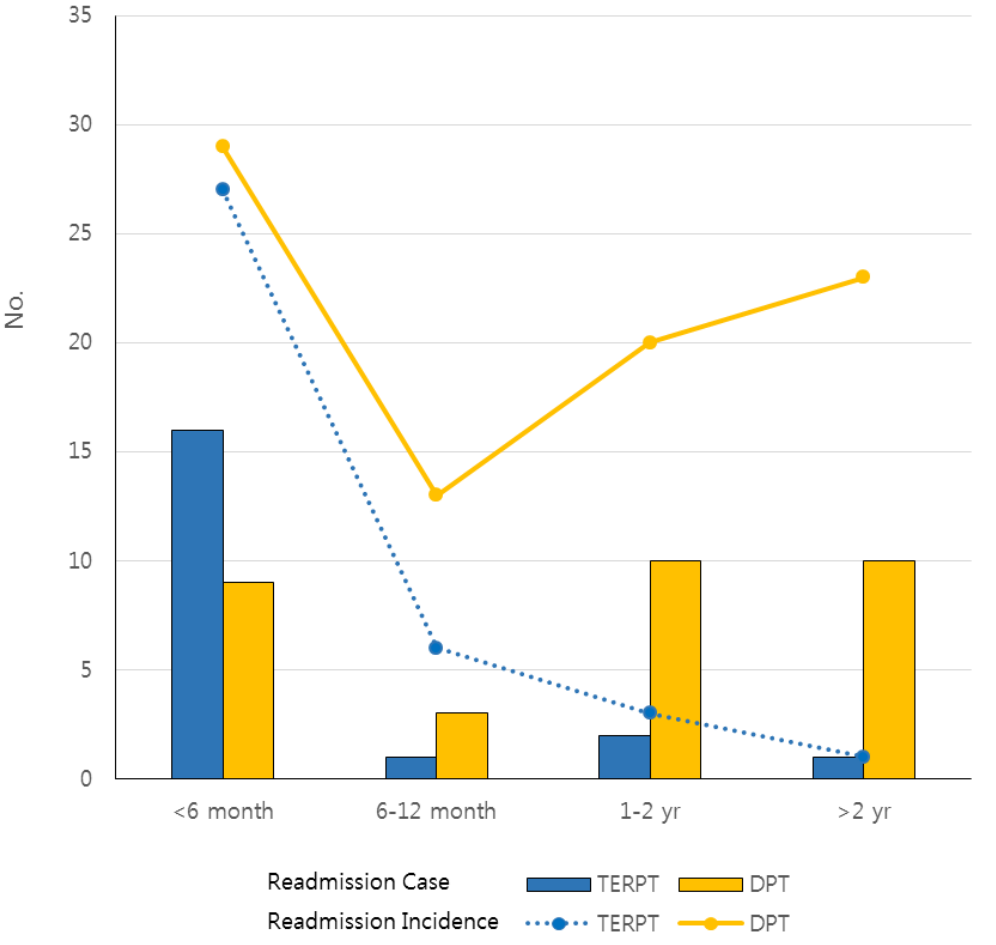

Figure 1. Readmission after operation

B. Re-operation

The TERPT group had 3 cases of reoperation. Two cases underwent anal myectomies because of unsatisfactory defecation, and they had excellent outcomes after reoperation. The other case underwent twice of anal myectomies and a BOTOX® injection, but his symptoms were not improved, and he underwent re-do TERPT.

In the DPT group, 23 cases underwent reoperations for a total of 44 times. This was significantly higher than in the TERPT group. The most common types of reoperations were anal myectomies and BOTOX® injections, and these

were followed by divisions of the septum in the anastomosis. The other types of reoperations included re-do pull-through, anal fistulotomy, colostomy and rectoplasty.

C. Functional outcomes

The functional outcomes were summarized in Table 5.

Forty-seven patients in the TERPT group and 37 patients in the DPT group could be followed beyond the age of 3. Functional outcomes of these patients were analyzed. There was no statistically significant difference in age between the two groups at final follow-up.

In the TERPT group, the functional outcome was excellent; most patients had no soiling or constipation. Only 2 cases had mild symptoms: one had grade 1 soiling, and the other case had both grade 1 soling and grade 1 constipation. In comparison, the DPT group showed a significantly higher incidence of soiling than the TERPT group; 16 cases (43.2%) had soiling, and 4 of them were grade 2. Constipation was also significantly more common in DPT group; 6 cases (16.2%) had constipation and 3 of them were higher than grade 1.

Table 5. Functional outcomes

	TERPT	DPT	P
Patients	47	37	
Mean age at follow-up (year)	7.1 ± 2.7	8.5 ± 3.9	0.056
Soiling			
No	45 (95.7)	21 (56.8)	<0.001
Yes	2 (4.3)	16 (43.2)	
Grade 1	2	12	
Grade 2	0	4	
Grade 3	0	0	
Constipation			
No	46 (97.9)	31 (83.8)	0.040
Yes	1 (2.1)	6 (16.2)	
Grade 1	1	3	
Grade 2	0	2	
Grade 3	0	1	

IV. DISCUSSION

Since the introduction of several different surgical methods treating Hirschsprung's disease, comparison of these method has been an interesting topic of studies. Before the introduction of TERPT, the comparison studies were mainly performed among the 3 transabdominal pull-through operations of Swenson, Soave, and Duhamel procedures.[9,10,20] TERPT was introduced in the late 1990s and has become a popular method for treating Hirschsprung's

disease.[12,13] Consequently, the comparison of the surgical outcomes between traditional operations and TERPT has become an important subject in Hirschsprung's disease. Among the three transabdominal pull-through operations, Duhamel procedure has become the one of popular methods and most recent studies have focused on the comparison between TERPT and DPT.[14,16,21-23]

The results of this study comparing the operative results and long term surgical outcomes of TERPT with those of DPT showed the superiority of the TERPT, not only in operative results but in long-term functional outcomes. Comparing with DPT, TERPT is a minimally invasive surgery which does not require an abdominal incision and wide intraabdominal manipulation. Abdominal dissection is minimal, and the risk of damage to the pelvic structure is also reduced. So operative time could be shortened and postoperative recovery is usually excellent. Recently TERPT has become the most commonly applied surgical method for the treatment of Hirschsprung's disease. The results of this study demonstrate that TERPT had significantly shorter operative times and fewer operation-related complications and these results are also compatible with previously published studies.[14,16,24,25]

However, unlike the operative outcomes, the functional outcomes of TERPT did not show a definite superiority over DPT. Recent studies have reported that TERPT had worse long-term functional outcomes than DPT or that there were no differences between the two procedures.[14,16,19,22] In this study, on the contrary, TERPT demonstrated significantly better outcomes than DPT. Postoperative re-admission and re-operation rates were significantly less than DPT. TERPT also showed the less soiling and less constipation than DPT in the follow-up period in this study.

The reasons why this study showed better outcomes over DPT are uncertain, but I think the differences in the procedures might play an important role. DPT was originally developed as a technical variation of the Swenson procedure to

avoid wide dissection of the pelvis and to preserve the important reflex area of the rectum.[7] DPT needs retrorectal dissection and preserving distal aganglionic rectum. Introduction of the GIA stapler and adaptation of a laparoscopic procedure are recent technical development of the procedures. On the other hand, TERPT was developed based on the Soave procedure. In the TERPT procedure, the aganglionic bowel could be removed completely, and endorectal dissection could minimize pelvic dissection and injury to pelvic structures.[8] Application of transanal dissection could minimize injuries to the anal sphincter and pelvic organs than the original dissection of the Soave procedure. Those differences could be interpreted as the factors that make TERPT superior.[21]

Although TERPT has the above-mentioned advantages, it also has a few well known disadvantages. Technically the anal sphincter needs to be overstretched because it is necessary to open the anus wide during TERPT. To avoid this technical pitfall, the surgeon who performed TERPT in this study used the colostomy plate and suture traction[26] and could reduce anal sphincter trauma with less retraction tensions.

The other consideration in this study is that the short cuff procedure was not performed. As one of the modifications of the classic TERPT, the short muscular cuff procedure had been introduced, in which transanal mucosal dissection is not required to reach the peritoneal cavity.[26,27] This procedure could avoid the stenotic rectum and accompanying obstructive symptoms produced by them, and the outcome has been better according to recent studies.[27,28] However, I think the long cuff procedure still has advantages. The risks of injuries to the adjacent pelvic structure are definitely low in the long cuff procedure. The ability to hold the pull-through colon is better than the short cuff procedure. Consequently, the chance of mucosal prolapse might be less than with the short cuff. Also, the dissection of the upper portion of the rectum is easier than that of the lower portion, so that it could be performed in short time. Enough of a split or partial excision of the cuff muscle could prevent the

postoperative obstructive symptoms that could be induced by the long cuff procedure.

The interesting finding in this study was the difference in postoperative readmission between two groups. In spite of the significantly higher total number of readmission cases with DPT, the number of readmission cases within 6 months after the operation were more common with TERPT, though the number of readmissions during this period was similar in the two groups. I think those results are due to the possible stabilization period after TERPT.[29] The ages of patients undergoing TERPT are usually the neonatal or early infant period, and these patients are easily affected by postoperative changes and they need a longer postoperative stabilization period. Therefore, TERPT showed a high incidence of readmissions during the 6 months after the operation.

In this study, only cases of aganglionosis confined to the rectosigmoid colon were analyzed. Although the majority of Hirschsprung's disease patients have the aganglionic bowel below the rectosigmoid, about one-fourth of patients have the long segment or total colonic aganglionosis.[17] The DPT is known to be a more favorable technique in patients with long segment Hirschsprung's disease.[21,30] Therefore, the comparison of the two operations with regard to long segment Hirschsprung's disease should be analyzed in further studies.

The other limitation of this study is the surgeon factor. In this study, all operations were performed by several pediatric surgeons, and results would be different depending on surgeons. The surgeons' preferences in postoperative management would be various depending on surgeons, so that delicately different indications of readmissions and reoperations would be applied. Those factors would be considered bias factors of which should be eliminated in future study.

V. CONCLUSION

TERPT showed shorter operative times, fewer operation-related complications, and fewer postoperative readmissions than DPT. Functional outcomes were also better in TERPT; the incidence of soiling and constipation were significantly lower in TERPT. These results support the superiority of TERPT over DPT.

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ABSTRACT (IN KOREAN)

히르슈슈프룽병에서의 one-stage transanal endorectal pull-through 술식과 Duhamel pull-through 술식의 비교

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차치환

목적 : 본 연구는 히르슈슈프룽병의 치료 술식인 one-stage transanal endorectal pull-through (TERPT)술식과 Duhamel pull-through (DPT)술식의 수술 결과 및 장기간 추적 후의 배변 기능을 비교하고자 하였다.

방법 : 2001년부터 2013년까지 1세 이전에 히르슈슈프룽병으로 세브란스 어린이 병원 소아외과에서 TERPT술식 또는 DPT술식으로 수술을 받은 환자들 중에서, 병변이 직장 구불결장에 국한된 환자를 대상으로 하였다. 후향적으로 환자의 임상 자료 및 수술 후 경과를 분석하였으며, 배변 기능의 평가는 수술 후 만 3세 이상인 환자를 대상으로 하였다.

결과 : 대상기간 동안 TERPT술식을 시행 받은 환자는 51명이었으며 DPT술식을 시행 받은 환자는 50명이었다. 환자들의 평균 수술 연령은 TERPT술식을 받은 환자들이 DPT술식을 받은 환자들보다 어렸으며(1.7 ± 1.9 vs. 4.0 ± 2.4 개월, p <0.001), 평균 수술 시간도 TERPT술식이 DPT술식보다 짧았다(154.6 ± 52.4 vs. 196.6 ± 65.0 분, p = 0.001). 수술 관련 합병증도 TERPT술식군이 DPT술식군보다 유의하게 적었으나(0 vs. 18 %, p = 0.001), 수술 후 입원기간은 TERPT술식군과 DPT술식군사이에 차이가 없었다(9.6 ± 3.1 vs. 11.4 ± 9.3 일, p = 0.200). 수술 후 재입원은 TERPT술식군에서 유의하게 낮았으며(39.2 vs. 64.0 %, p = 0.013), 가장 흔한 재입원 이유는 두 군 모두 결장염이었다. 재입원은 TERPT술식군에서는 재입원한 환자의 80%에서 수술 후 6개월 이내에 재입원이 발생하였으나, DPT술식군에서는 재입원한 환자의 62.5%에서 수술 후 1년이 지나서 재입원이 발생하였다. 장기간 추적 후의 배변 기능은 TERPT술식군이

DPT술식군보다 변실금 (4.3 vs. 43.2 %, p <0.001)과 변비 (2.1 vs. 16.2 %, p = 0.040)의 발생률이 유의하게 낮았다.

결론 : 본 연구는 TERPT술식군이 DPT술식군보다 수술 후 임상결과가 유의하게 우수함을 보여 주었다. 이러한 결과는 히르슈슈프룽병의 치료 술식으로 DPT술식보다 TERPT술식이 더 적합하다는 것을 뒷받침한다.

핵심되는 말 : 히르슈슈프룽병, transanal endorectal pull-through술식, Duhamel술식