

Original Article

Long term outcomes for neonates of Hirschsprung's disease undergoing transanal Swenson or Duhamel pull-through by a 5 year follow-up study

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Abstract: Objective: To study long term outcomes for neonates of Hirschsprung's disease undergoing transanal Swenson or Duhamel pull-through methods. Methods: In the present study, a retrospective analysis was conducted including 79 HD neonatal cases who received transanal Soave or Duhamel pull-through surgery from January 2001 to December 2011. Among all the patients, 29 underwent the transanal Swenson procedure and 50 underwent the Duhamel pull-through procedure. Data including basic demographics, pathological type, operation type, operative time, blood loss, hospital stay, bowel function, and complications were collected from all patients. A 5 year follow-up for all patients and information of postoperative complications and bowel function were recorded at 3 months, 6 months, 2 years, and 5 years after the operation respectively. Results: The operation time and hospital stay of patients in the Swenson group were significantly lower than those of the patients in the Duhamel group with $P < 0.05$, while other clinical outcomes showed no significant difference. Both intra-operative complications and short-term post-operative complications evaluated 3 months after the surgery had no significant difference between the two groups, $P > 0.05$. The complication rate decreased in all patients 2 years after surgery. However, complication rates of the Swenson group during 6 months to 5 years were all higher than those of the Duhamel group, and the difference was significant at the follow-up period of 5 years with $P < 0.05$. Results of Rintala score didn't show a significant difference between the two groups in the 5 years of follow-up. However, the Rintala score of the Duhamel group was higher during all follow-up periods. Conclusion: The transanal Swenson method may have a shorter operation time and hospital stay, however the Duhamel pull-through method may have a better long term outcome. This study can provide more clinical evidence and better understanding for the treatment of HD patients.

Keywords: Hirschsprung's disease, follow-up, transanal Swenson, Duhamel, pull-through

Introduction

Hirschsprung's disease (HD) is a rare congenital disease characterized by neonatal functional low bowel obstruction that affects almost 2% of the people all over the world [1]. HD results in secondary absence of ganglion cells in the distal region of the intestinal wall and leads to serious developmental disorders of the alimentary system [2]. Studies show that in most cases, HD appears in newborns or early infancy, which increases the difficulty of treatment. There are also cases presenting late into childhood, adolescence, or even in adults, which is still a challenge to the clinic [3-5].

Since the first reported operative approach for HD by Orvar Swenson in 1948 [6], more and

more patients benefit from the development of surgery techniques and deeper understanding of the pathologic anatomy and physiology of the defect with better prognosis and higher survival rates. At present, surgery is the most common method for treatment of HD such as the endorectal dissection (Soave) [7] and retrorectal procedure (Duhamel) [8]. The development of a single-stage pull-through strategy is one of the most important developments for surgical management of HD in the last 3 decades, and has gradually replaced the traditional 2 or 3 stage procedure [9].

Among these strategies, the transanal Swenson method and Duhamel procedures are two of the latest methods adopted in the clinic and are becoming increasingly popular [10, 11].

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Table 1. Basic clinical information of the patients

	Swenson, n = 29	Duhamel n = 50	Mean	P value*
Age at the time of operation, years	2.13 ± 0.42	2.29 ± 0.61	2.20 ± 0.56	0.643
Weight, kg	10.39 ± 0.64	10.76 ± 0.85	10.48 ± 0.44	0.729
Type				
Short segment (proximal and middle rectum)	14	25		
Ordinary (rectosigmoid)	10	17		0.958
Long segment (sigmoid and descending)	5	8		

*Comparison between Swenson and Duhamel.

Transanal Swenson pull-through operation is the earliest modification of transanal approach which is considered to be a combination of transanal Soave method and transabdominal Swenson procedure that completely remove the aganglionic bowel [12]. Duhamel pull-through procedure was first reported in 1994 and has been modified through the years to reduce the size of the surgical wound, minimize injury to surrounding structures during intra-abdominal dissection, and improve bowel function [13]. Compared to traditional approaches, the newly developed pull-through procedures have lower hospital stays and fewer postoperative complications [14]. However, the long term outcomes of these surgical approaches are less reported [15].

To the best of our knowledge, comparison of long term outcomes for HD patients undergoing transanal Soave or Duhamel pull-through is rarely reported and clinical data for the long term prognosis of the two procedures for treatment of neonates are still inadequate. In the present study, we report a five-year follow-up study for 19 HD neonates who have received transanal Soave or Duhamel pull-through surgery in our hospital. This study will give more clinical evidence and better understanding for the long term effects of transanal Soave and Duhamel pull-through procedures in treatment for HD neonates.

Materials and methods

Patients

In the present study, a retrospective analysis was conducted including 79 HD neonatal cases who received transanal Soave or Duhamel pull-through surgery in Jining First People's Hospital from January 2001 to December 2011. Among all patients, 29 (mean age 2.13 ± 0.42 , male:female = 15:14) underwent the

transanal Swenson procedure and 50 (mean age 2.29 ± 0.61 , male:female = 26:24) underwent the Duhamel pull-through procedure. Symptoms of intractable constipation and abdominal distention were diagnosed in all patients, and glycerine enema was used to release the patients' symptoms. Diagnosis was confirmed by barium enema and rectal mucosa biopsy. Patients who had laparoscopic operations were excluded from the study. Detailed clinical information of all patients was listed in **Table 1**. Informed consents were obtained from all patients and the present study was approved by the Ethics Committee of Jining First People's Hospital.

Surgery

Pre-operation preparation included repeat cleansing enema for 5~20 days (median 7 days), and bowel preparation was performed 3 days before surgery, including improving the liquid, taking oral gentamicin, metronidazole and vitamin K4. The Swenson procedure was performed according to Sookpotarom's report [12] and the open Duhamel procedure was reported elsewhere [16]. The principle of the surgery strategy was the same in all patients undergoing the same procedure.

Data collection and follow-up

Data including basic demographics such as age and weight at the time of operation, pathological type, operation type, operative time, blood loss, hospital stay, bowel function, and complications were collected from all patients. The Rintala scale was used to evaluate the bowel function recovery [17]. A 5 year follow-up was done for all patients at the outpatient clinics or by telephone. Information on postoperative complications and bowel function were recorded at 3 months, 6 months, 2 years, and 5 years after the operation respectively.

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Table 2. Clinical outcomes for patients with different procedures

	Swenson, n = 29	Duhamel, n = 50	Mean	P value*
Operation time, min	105.65 ± 28.72	121.32 ± 32.43	110.25 ± 32.44	0.036
Blood loss, ml	23.45 ± 10.12	25.66 ± 9.31	24.15 ± 11.65	0.876
hospital stay, day	7.4 ± 2.3	11.3 ± 2.2	9.1 ± 2.5	0.021
Rintala score#	15.46 ± 4.28	16.19 ± 6.32	15.84 ± 5.13	0.355
Intra operative complications				
Urethral injury	1	1		
Bowel injury	1	1		
Vaginal injury	1	1		
Aganglionic bowel left behind	0	1		
Total	3 (10.3%)	4 (8.0%)		0.621
Postoperative complications#				
Fecal soiling	1	2		
Anastomotic fistula	1	2		
Enterocolitis	1	1		
Recurrent of constipation	1	1		
Anastomotic stenosis	1	1		
Total colonic prolapse	0	1		
Total	5 (17.2%)	8 (16.0%)		0.849

*Comparison between Swenson and Duhamel. #Evaluated 3 months after surgery.

Statistical analysis

Measurement data was expressed by the mean ± SD. Independent continuous variables were compared using the Student *t*-test and categorical data were compared using the Chi square test or Fisher exact tests. It was considered to be statistically significant when a *P*-value was less than 0.05. All calculations were made using SPSS 18.0.

Results

Basic demographics of the patients

In the present study, a total of 79 neonatal patients were included, with 29 patients undergoing Swenson procedure and 50 patients undergoing Duhamel pull-through procedure. As shown in **Table 1**, the age, weight, and pathological type of patients in different surgical groups had no significant difference, *P*>0.05. All patients were divided into 3 groups of short segment HD (defined as those with aganglionosis extending to proximal and middle rectum), ordinary HD (defined as those with aganglionosis extending to rectosigmoid), and long segment HD (defined as those with aganglionosis extending to sigmoid and descending) for each surgery procedure. The basic clinical information of all patients is listed in **Table 1**.

Clinical outcomes for patients with different procedures

Clinical outcomes and short term complications for patients in different surgical groups are listed in **Table 2**. For clinical outcomes, the operation time, blood loss, hospital stay, and bowel function were collected and analyzed. Results show that the operation time and hospital stay of patients in Swenson group are significantly lower than those of the patients in Duhamel group, *P*<0.05, while other clinical outcomes show no significant difference between the two groups. Analysis of short term complications of the two surgeries shows that both intra-operative complications and short-term post-operative complications evaluated 3 months after the surgery, had no significant difference between the two groups, *P*>0.05.

Complications and bowel function of the patients in a five-year follow-up

Data of the complications and bowel function of all patients were collected in a five-year follow-up at 3 months, 6 months, 2 years, and 5 years after surgery respectively. As shown in **Table 3**, the complication rate decreased in all patients in 2 years after surgery. However, except for the complication rate at 3 months, complication rates of Swenson group during 6

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Table 3. Long term results of complications and bowel function of the patients in a five-year follow-up

	3 months	6 months	2 years	5 years
Complication, cases				
Swenson, n = 29	5 (17.2%)	4 (13.8%)	3 (10.3%)	4 (13.8%)
Duhamel, n = 50	8 (16.0%)	4 (8.0%)	2 (4.0%)	2 (4.0%)*
Rintala score				
Swenson, n = 29	15.46 ± 4.28	15.78 ± 4.11	15.64 ± 3.18	15.27 ± 5.35
Duhamel, n = 50	16.19 ± 6.32	16.53 ± 5.12	16.56 ± 3.65	16.29 ± 3.23

*P<0.05, compared with the Swenson group.

months to 5 years were all higher than those of the Duhamel group, and the difference was significant at the follow-up period of 5 years, with P<0.05, indicating that the Duhamel procedure might have better long term outcomes. Three cases reported a disturbance of defecation and 2 cases got enterocolitis in the Swenson group after 6 months. While 4 cases had recurrent constipation and 4 cases had fecal soiling in the Duhamel group, respectively after 6 months. After 2 years, 2 cases had symptoms of defecation dysfunction and 1 case had reoccurrence in the Swenson group, and 2 cases had defecation dysfunction in the Duhamel group. After 5 years, 4 cases had symptoms of defecation dysfunction in the Swenson group while 2 cases had the same symptoms in the Duhamel group. Results of the Rintala score didn't show significant difference between the two groups, however the Rintala score of the Duhamel group was higher at all follow-up periods.

Discussion

In recent years, development of surgical approaches have provided benefit to more and more Hirschsprung's disease (HD) patients. However, insufficiency and complications still exist in many cases. Studies show that inappropriate treatment may lead to a risk of life-threatening enterocolitis in neonates or failure to thrive with defecation problems in older children [18]. Both transanal Swenson and Duhamel pull-through approaches are the newest pull-through surgery strategies which are reported to have better outcomes and fewer complications than old traditional methods. However, some studies also reported complications were found in cases undergoing the above two surgical procedures.

In a review study for long term (most more than 10 years) living conditions for HD patients, it was reported that they might have uniformly lower scores of overall bowel function than healthy control subjects and functional outcome and quality of life may be interrelated and deteri-

orate with increasing age [19]. Chiengkriwate et al. reported that 4 (13.3%) of 30 HD patients undergoing primary pull-through with a modified Duhamel technique had acute postoperative complications [20]. In a comparison study between the Swenson and the Soave procedures for HD patients, both of the two methods showed complication rates of more than 15% [21]. All these studies indicate that surgical approaches adopted presently may not be sufficient enough and thus more studies should be done to provide more clinical basis and evidence for understanding HD surgical methods.

To the best of our knowledge, fewer studies have focused on comparison between short and long term outcomes of HD patients undergoing transanal Swenson or Duhamel pull-through surgeries. Thus in the present study, we tried to conducted a retrospective analysis for HD patients with two surgical methods by a 5-year follow-up.

In this study, we found that patients with the Swenson procedure had significantly shorter operation time and hospital stay compared with those of the patients in Duhamel group, while other clinical outcomes showed no significant difference between the two groups. Complication analysis showed that short term complications of the two surgeries were not significantly different. However, complication rates of the Swenson group during 6 months to 5 years were all higher than those of the Duhamel group, and the difference was significant at the follow-up period of 5 years, suggesting that patients with the Duhamel method might have better long term outcomes comparing with the Swenson method.

Some related studies were reported. Tannuri et al. reported that long-term results of the

Duhamel technique are superior to those of the transanal pull-through, which is partly in consistent with our study [22]. A comparison study of open and laparoscopic Duhamel pull-through techniques showed that both of the two methods had similar outcomes [16]. An older research comparing Soave's and Duhamel's pull-through methods showed that there was no significant difference between the two procedures in postoperative surgical morbidity or in long-term risk of enterocolitis [23]. Arts et al. reviewed Duhamel and transanal endorectal pull through (TERPT) for the surgical treatment of Hirschsprung's disease and found that evidence was insufficient to recommend one technique over the other and the surgeon's experience might be the key factor determining the choice of procedure [24].

In conclusion, we conducted a retrospective analysis to study long term outcomes of HD patients undergoing transanal Swenson or Duhamel pull-through methods. Results show that the transanal Swenson method may have a shorter operation time and hospital stay, however the Duhamel pull-through method may have better long term outcomes. This study thus provides more clinical evidence and better understanding for the treatment of HD patients.

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Disclosure of conflict of interest

None.

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