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Original Article

Results of the Treatment of Hirschsprung's Disease: Comparison between Transabdominal and Transanal Endorectal Pull-through Operations

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Abstract

Background: **Transabdominal pull-through operation has been established as the definitive treatment of Hirschsprung's disease (HD) since 1948. One-stage transanal endorectal pull-through procedure is the latest evolution in the management of HD.**

Objective: **The aim of this study was to review the results of the treatment of HD by comparing between transabdomial and transanal endorectal pull-through operations.**

Materials and Methods: **Medical records of patients with HD who underwent definitive treatment between January 2007 and December 2012 at Queen Sirikit National Institute of Child Health were retrospectively reviewed. Patients who underwent transabdominal and transanal endorectal pull-through operations were categorized into groups A and B, respectively. Results of the two groups were compared using the Chi-square test, and a p-value less than 0.05 was considered statistically significant.**

Results: **Of the 145 patients with HD, 86 (59%) were treated by transbdominal pull-through operation (group A), and 59 (41%) were treated by transanal pull-through procedure (group B). Age at operation ranged from 1.6 months to 12.7 years in group A and 21 days to 5.6 years in group B. One year after operation, normal defecation and continence were noted in 71% of patients in group A, and 70% in group B ($p = 0.925$). Constipation was more common in group A than that in group B (15% vs. 7%; $p = 0.032$). Anastomotic leakage (4%) and adhesive small bowel obstruction (4%) occurred in group A, but did not occur in group B. Only anastomotic stricture which required anal dilatation was more common in group B than that in group A, but with no statistical significance (9% vs. 4%; $p = 0.267$). The incidence of postoperative enterocolitis was not different between groups A (20%) and B (20%). There was no immediate postoperative death in both groups.**

Conclusions: **Operative outcomes from the present study revealed that transanal endorectal pull-through operation was better than the transabdominal procedure in terms of the low incidence of constipation, anastomotic leakage, postoperative small bowel obstruction and constipation. Occurrence of normal defecation and the incidence of postoperative enterocolitis were not different between the two techniques.**

Keywords: **Hirschsprung's disease, abdominal pull-through operation, transanal endorectal pull-through, operation, results of treatment, enterocolitis, constipation**

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Introduction

Hirschsprung's disease, or "congenital megacolon", was first described by Harald Hirschsprung, a pathologist at Queen Louise Children's Hospital in Copenhagen, in a report of two cases in 1887[1]. Patients with HD often present with the symptoms of functional intestinal obstruction due to the absence of ganglion cells in the distal colonic wall, most commonly at the rectum and sigmoid colon. The obstruction develops at the level of colonic aganglionosis. The aganglionic segment may extend more proximally into the descending and transverse colon. Sometimes the aganglionosis involves the entire colon and even the terminal ileum, which is called total colonic aganglionosis. The three most common operative techniques for definite treatment of HD include the Swenson (1948)[2], Duhamel (1956)[3] and Soave procedures (1963)[4]. These procedures are performed via a transabdominal approach, and has been refined in recent years with fewer associated complications.

The one-stage transanal endorectal pull-through procedure (TEPT) is the latest in the evolution of management of HD, first described by De la Torre-Mondragon and Ortega-Salgato in 1998[5]. They first described the totally TEPT procedure in neonates. TEPT is currently popular and is claimed to be less costly, with lower complications than the previous abdominal pull-through operations (APTs). Therefore, the authors retrospectively reviewed the experience in the management of HD at a tertiary institute for pediatrics. The aim of this study was to compare results of the treatment of HD between APT and TEPT, in a recent 6-year period.

Materials and Methods

Medical records of patients with HD treated with definitive surgery at Queen Sirikit National Institute of Child Health between January 2007 and December 2012 were reviewed. The study was begun after the approval by the Institutional Review Board (Document No. 57-023). Patients who underwent APT (Swenson, Duhamel and Soave procedures) and TEPT were labeled as groups A and B, respectively. Results of treatment of the two groups were compared using the Chi-square test. A p-value less than 0.005 was considered statistically significant. The study focused only on HD with absence of ganglion cells within some parts of the colon. Patients with total colonic aganglionosis were excluded from the study.

Results

During the study period, 145 patients were treated for HD with definitive surgery, with 86 (59%) treated by APT (group A) and 57 (41%) by TEPT (group B) (Figure 1). Clinical characteristics were not significantly different between the two groups, except age at clinical presentation and age at definitive operation (Table 1). Patients in group A were older than those in group B (mean age at clinical presentation, 6.4 ± 34.1 vs 1.7 ± 9.2 months, $p < 0.001$ and mean age at pull-through operation, 18.8 ± 96.7 vs 7.9 ± 25.5 months, $p = 0.029$).

In group A, the transitional zone was at the rectosigmoid level in 27 of 86 cases (31%) and in group B, 36 of 59 cases (61%), which was statistically different ($p < 0.001$). Before APT, patients in group A had primary colostomy in 59 cases (69%), while all of the 59 patients in group B did not undergo colostomy before TEPT. The average duration of operation was significantly higher in group A (4.5 ± 1.3 hrsvs 3.0 ± 1.3 hrs, $p = 0.010$).

Postoperative complications between the two groups were not significantly different, except wound infection, which was more common in group A (15 % vs 3 %, $p = 0.027$), see Table 2. Results of treatment at one-year follow-up after definitive pull-through operations revealed that 70% of patients in both groups had normal defecation (Table 3). However, patients

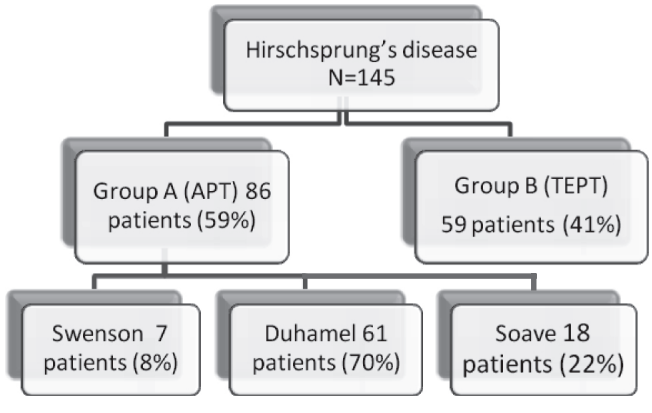

Figure 1 Schematic diagram for definitive treatment of the 145 patients with Hirschsprung's disease, group A. abdominal pull-through operation, group B. transanal endorectal pull-through operation

Table 1 Clinical characteristics of patients in both groups

Demographic data	Group A APT (n=86)	Group B TEPT (n=59)	*p*-value
Gender			
Male: female	68:18 (3.7:1)	53:6 (8.8:1)	0.112
Gestational age (wks)	31-42	30-42	
Birth weight (gms)	1170-4000 (2946 ± 940)	1340-4214 (3048 ± 1066)	0.433
Age at clinical presentations	2 days-103 mo (6.4 ± 34.1 mo)	3 days-35 mo(1.7 ± 9.2 mo)	<0.001
Preoperative enterocolitis	27 (31.4%)	16 (27.1%)	0.194
Age at definitive operation	1.6 mo-12yrs (18.1 ± 51.0 mo)	21 days-5.6yrs (7.9 ± 25.5mo)	<0.001
Operative time	3.55-5.4 hrs (4.50 ± 1.3 hrs)	2.30- 4.4 hrs (3.04 ± 1.3 hrs)	0.010

Table 2 Postoperative complications in group A and group B

Complications (cases/percentage)	Group A APT (n=86)	Group B TEPT (n=59)	*p*-value
Wound infection	13 (15.1)	2 (3.4)	0.027*
Anastomosis leakage	3 (3.5)	0	0.274
Gut obstruction	3 (3.5)	0	0.274
Anastomotic stricture	3 (3.5)	5 (8.5)	0.267
Enterocolitis	17 (19.8)	12 (20.3)	1.000
Anastomotic dysfunction	0	1 (1.7)	0.401
Perianal abscess	1 (1.2)	0	0.401
Wound dehiscence	1 (1.2)	0	0.401

Table 3 Evaluation at the first year after operation

Clinical outcomes (cases/percentage)	Group A APT (n=86)	Group B TEPT (n=59)	*p*-value
Normal defecation (1-3 times/day)	61 (70.9)	41 (69.5)	0.925
Constipation	13 (15.1)	4 (6.8)	0.032*
Frequency of defecation> 3 times/day	12 (14.0)	14 (23.7)	0.667

in group A developed a significantly higher frequency of constipation than those in group B (15 % vs 7 %, $p = 0.032$), while diarrhea was more frequent in group B, but with no statistical significance (24 % vs 14 %, $p = 0.667$).

Discussion

During the period between 1950 and 2000, patients with HD were treated by temporary colostomy and transabdominal pull-through operation in most cases. Three types of operations, which included the Swenson, Duhamel and Soave procedures, were performed at our institute. Long-term outcomes were evaluated in 2008, and showed satisfactory results, according to the quality of life at 5 to 25 years after operation[6]. Our experience with each type of APT procedures was similar to those in previous reports[7-13]. However, since APT procedures were usually not done in neonates and young infants, some patients required temporary colostomy in order to decrease rectal irrigation and prevent enterocolitis before definitive pull-through operations.

After De la Torre-Mondragon and Ortega-Salgado[5] reported TEPT in 1998, this technique has gained world-wide popularity, in HD with low-segment

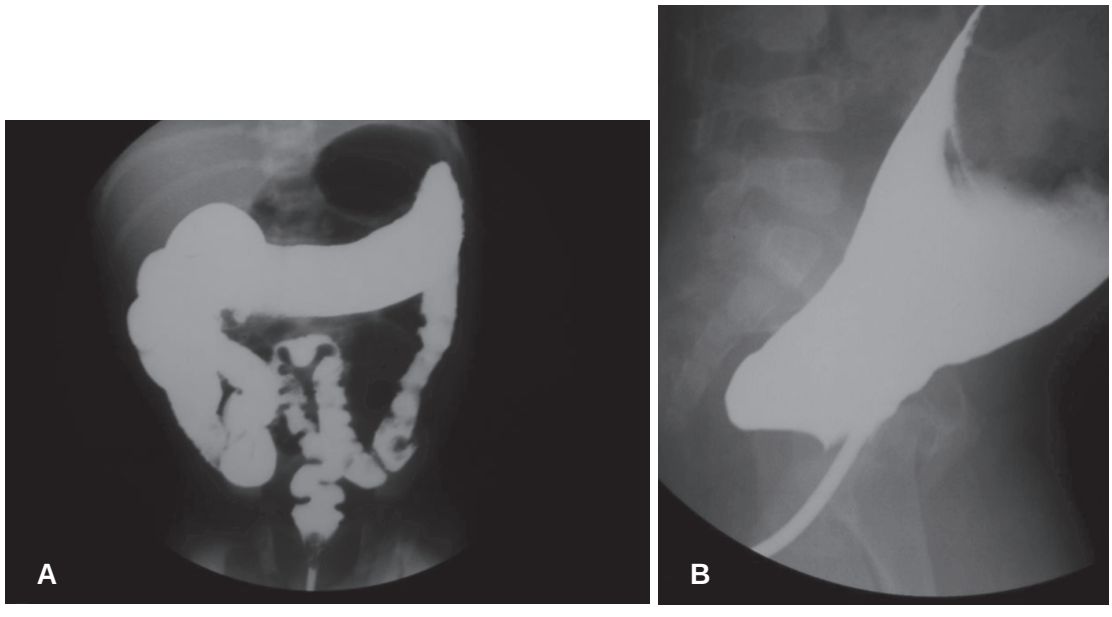

Figure 2 Level of aganalionosis A. at the splenic flexure and treated by Duhamel's pull-through operation, B. at the rectosigmoid colon treated by TEPT

aganglionosis. Since then, many investigators reported their experience with TEPT for HD, with favorable results[14-18]. Studies comparing APT and TEPT were published by many investigators[19-23], and TEPT was generally found to be more cosmetic, with less postoperative pain, surgical complications, shorter hospital stay, and reduced cost. The results of a retrospective review from our institute in 2011, of 53 patients with HD treated by TEPT, were similar to those or other reports but a comparison with open abdominal pull-through procedure was not done[23].

In the present study, we compared clinical outcomes between APT and TEPT and confirmed that TEPT yielded better results, in terms of shorter operative time, low incidence of wound infection, anastomotic leakage and postoperative gut obstruction, in addition to being applicable to neonates. The disadvantages of TEPT included postoperative anastomotic stricture, and TEPT could not be done when the aganglionosis was higher than the sigmoid colon. The more common complications of TEPT reported in the literature included anastomotic stricture, constipation, and enterocolitis[24,25]. In contrast, in present study TEPT was associated with less constipation than APT, whereas normal defecation and the incidence of postoperative enterocolitis were similar to those of APT.

Conclusion

Operative outcomes of definitive treatment for HD in the present study revealed that TEPT was superior to APT procedure in terms of wound infection, anastomotic leakage, postoperative small bowel obstruction and low incidence of constipation. Disadvantages of TEPT included anastomotic stricture and the limitation to cases with aganglionosis located higher than the sigmoid colon. Normal defecation and the incidence of postoperative enterocolitis were not different between the two procedures.

Acknowledgement

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บทคัดย่อ **ผลผลการรักษาโรคไม่มีเซลล์ปมประสาทในผนังของลำไส้ใหญ่: เปรียบเทียบระหว่างการผ่าตัดทางช่องท้องกับการผ่าตัดทางทวาร**

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**กลุ่มงานศัลยศาสตร์ สถาบันสุขภาพเด็กแห่งชาติมหาราชินี กรุงเทพฯ*

***กลุ่มงานรังสีวิทยา สถาบันสุขภาพเด็กแห่งชาติมหาราชินี กรุงเทพฯ*

ความเป็นมา: การผ่าตัดเปิดหน้าท้องเพื่อดึงลำไส้ส่วนที่มีเซลล์ปมประสาทลงไปต่อที่ทวาร ในการรักษาโรคไม่มีเซลล์ปมประสาทในผนังลำไส้ใหญ่ เริ่มมาตั้งแต่ปี ค.ศ. 1948 การผ่าตัดแก้ไขทางทวารเป็นวิวัฒนาการล่าสุดในการรักษาโรคนี้

วัตถุประสงค์: เพื่อศึกษาถึงผลของการรักษาโรคไม่มีเซลล์ปมประสาทในผนังของลำไส้ใหญ่ โดยการเปรียบเทียบกันระหว่างการผ่าตัดเปิดหน้าท้องกับการผ่าตัดทางทวาร

วัสดุและวิธีการ: เวชระเบียนของผู้ป่วยที่เป็นโรคไม่เซลล์ปมประสาทในผนังของลำไส้ใหญ่ ซึ่งเข้ารับการผ่าตัดรักษาโรคนี้ระหว่างเดือนมกราคม ค.ศ. 2007 ถึงเดือนธันวาคม ค.ศ. 2012 ได้ถูกนำมาศึกษาย้อนหลัง ผู้ป่วยที่ผ่าตัดเปิดหน้าท้องถูกจัดให้เป็นกลุ่ม A ส่วนผู้ป่วยที่ผ่าตัดแก้ไขทางทวารถูกจัดเป็นกลุ่ม B ผลของการรักษาทั้งสองกลุ่มนำมาเปรียบเทียบกันโดยใช้ Chi-square test และค่า p น้อยกว่า 0.05 แสดงว่ามีความแตกต่างกันอย่างมีนัยสำคัญทางสถิติ

ผล: มีผู้ป่วยเป็นโรคไม่มีเซลล์ปมประสาทในผนังของลำไส้ใหญ่ที่นำมาศึกษาทั้งหมด 145 ราย 86 ราย (ร้อยละ 51) ได้รับการผ่าตัดทางช่องท้อง (กลุ่ม A) และ 59 ราย (ร้อยละ41) ได้รับการผ่าตัดทางทวาร (กลุ่ม B) อายุที่ทำการผ่าตัดอยู่ระหว่าง 1.6 เดือน ถึง 12.7 ปี ในกลุ่ม A และระหว่าง 21 วัน ถึง 5.6 ปีในกลุ่ม B หนึ่งปีภายหลังการผ่าตัดพบว่าถ่ายอุจจาระได้ปกติและกลั้นอุจจาระได้ร้อยละ 71 ของผู้ป่วยในกลุ่ม A และร้อยละ 70 ในกลุ่ม B (p=0.925) อาการท้องผูกพบในกลุ่ม A มากกว่ากลุ่ม B (ร้อยละ 15: ร้อยละ 7, p=0.32) การรั่วของลำไส้ที่เย็บต่อ (ร้อยละ 4) และลำไส้อุดตันหลังการผ่าตัดจากเยื่อพังผืด (ร้อยละ 4) พบในผู้ป่วยกลุ่ม A แต่ไม่พบภาวะแทรกซ้อนเหล่านี้ในกลุ่ม B มีเพียงการตีบของรอยเย็บต่อที่ทวารหนัก ซึ่งต้องรักษาโดยการถ่างขยาย พบในกลุ่ม B มากกว่ากลุ่ม A และไม่มีความแตกต่างกันอย่างมีนัยสำคัญทางสถิติ (ร้อยละ 9 : ร้อยละ 4, p=0.267) อุบัติการณ์ของการเกิดลำไส้อักเสบรุนแรงหลังการผ่าตัด ไม่มีความแตกต่างกันระหว่างกลุ่ม A (ร้อยละ 20) และกลุ่ม B (ร้อยละ 20) ไม่มีผู้ป่วยเสียชีวิตในระยะแรกหลังการผ่าตัดในผู้ป่วยทั้งสองกลุ่ม

สรุป: ผลของการผ่าตัดในการศึกษาครั้งนี้พบว่าการผ่าตัดผ่านทางทวารในการดึงเอาลำไส้ใหญ่ส่วนมีเซลล์ปมประสาทลงไปต่อบริเวณทวารหนักได้ผลดีกว่า การผ่าตัดผ่านช่องท้องในเรื่องของอัตราการเกิดการรั่วของรอยเย็บต่อลำไส้ การเกิดลำไส้อุดตันจากเยื่อพังผืดหลังผ่าตัด และอาการท้องผูกพบว่าอัตราการถ่ายอุจจาระได้ปกติและอัตราการเกิดลำไส้อักเสบที่รุนแรงหลังผ่าตัดไม่มีความแตกต่างกันระหว่างเทคนิคการผ่าตัดทั้งสองวิธี