

Original Research Article

Transanal endorectal pull-through in children as the treatment for Hirschsprung's in Aceh, Indonesia

Muntadhar M. Isa^{1,2}, Dian A. Syahputra^{1,2}, Muhammad Bayu Z. Hutagalung^{2*}

¹Pediatric Surgery Division, ²Department of Surgery, Faculty of Medicine, Syiah Kuala University/Dr. Zainoel Abidin Hospital, Banda Aceh, Aceh, Indonesia

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***Correspondence:**

Dr. Muhammad Bayu Z. Hutagalung,
E-mail: bayuzohari@gmail.com

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ABSTRACT

Background: One-stage pull-through operation has become increasingly popular for the treatment of Hirschsprung's disease. The advantages of total transanal pull-through include minimal resection of the dilated ganglionic part of the colon, shorter hospital stay, decreased total cost, lower risk of adhesive intestinal obstruction.

Methods: A retrospective study of patients with HD underwent transanal endorectal pull-through (TERPT) procedure treated at Dr. Zainoel Abidin Hospital Banda Aceh, Indonesia between January 2010 – December 2013. We assessed patients characteristic, outcome and complication including Hirschsprung associated enterocolitis (HAEC).

Results: A total 77 patients were included in this study. The mean age was 13.01 months (range from 11 days – 8 years old). Most of the patients (55.8%) were male and female (44.2%). Clinical classification of HD: short segment 74 patients (96.1%) and ultra-short segment 3 patients (3.9%). The mean of bowel resection length in TERPT procedure was 18.64 cm (range from 7–25 cm). There is no mortality associated with TERPT procedure. Hirschsprung associated enterocolitis (HAEC) occurred in 43 subjects (54.5%). Statistical analysis showed the significant relation between age at surgery and HAEC ($p=0.000$) and no significant relationship between gender ($p=0.425$) and bowel resection length ($p=0.780$) with HAEC.

Conclusions: Transanal endorectal pull-through procedure has been shown as an effective minimally invasive treatment in resolving obstructive symptoms in ultra-short segment of HD patients. The number of HAEC incidence among HD patients underwent TERPT significantly increased with older age of children.

Keywords: Transanal endorectal pull-through, Hirschsprung's disease, Treatment

INTRODUCTION

Hirschsprung's disease (HD) is a congenital bowel motility disorder with an incidence of 1:7000 to 1:4000 live born in the western world. Hirschsprung's disease patients usually present with bowel obstruction in the early neonatal period. Hirschsprung's disease can also be diagnosed in older children or adults with the history of recurrent constipation. Hirschsprung associated enterocolitis (HAEC) may also be the presenting symptoms.^{1,2}

The removal of the pathologic aganglionic bowel segment and reestablish bowel continuity are the main principle of treatment in HD. The minimally invasive surgical techniques have increased in popularity in these past decades. This led to a number of modifications of the standard HD operations, like the transanal endorectal pull-through, a modified Soave procedure with the endorectal dissection performed from below. There has also been a shift from three stage towards one stage surgery in the neonatal period without a diverting stoma.³

The advantages of total transanal pull-through include minimal resection of the dilated ganglionic part, decreased the length of hospital stay, decreased total cost, lower risk of adhesive intestinal obstruction. Hirschsprung associated enterocolitis is a complication of HD which can occur before and/or after definitive treatment. This condition characterized by abdominal distension, diarrhea and fever, and the exact pathogenesis is still unknown. Several patients have episodes of HEC after surgery. The outcome complications from this procedure vary around the world and it's believed to be associated with various factors such as age, gender and length of aganglionic segment.¹⁻³

METHODS

A retrospective study of patients with HD underwent Transanal Endorectal Pull-through (TERPT) procedure treated at dr. Zainoel Abidin Hospital Banda Aceh, Indonesia between January 2010 – December 2013. Diagnosis is made by a combination of medical history, examination, a barium enema to determine the type of HD using the level of the transition zone and rectal biopsy. The feasibility of a TEPT was assessed by a preoperative barium enema and the patient general condition. The primary transanal procedure is prepared if the radiological transition zone appeared in the rectosigmoid or mid-sigmoid region. The rectal biopsy is used to give a definitive diagnosis, performed at least 1 cm above the pectinate line and types of biopsies were submucosal biopsies and full thickness muscle coat stained with hematoxylin and eosin.

The operative technique was adopted from the combination of different investigators to obtain the best results. The patient was placed in a supine position, and a colostomy ring was placed around the anal outlet as an anal retractor. Eight deep stitches incorporating the anal sphincters were taken through the anal crypts and tied to the colostomy ring. A circumferential incision was made 5 mm above the dentate line. Submucosal dissection was carried out for a distance of 3 to 4 cm proximally. At this level, the muscular sleeve could be pulled downward, indicating that the dissection had reached a level that was well above the muscular pelvic floor. When the submucosal dissection extended proximally to a point above the peritoneal reflection, the rectal muscle was divided circumferentially, and the full thickness of rectum and sigmoid mobilized out through the anus. The colon was then divided several centimeters above the most proximal normal biopsy site, and a standard Soave-Boley anastomosis was performed. Oral feeding was started on the first postoperative day.^{4,5}

The subject characteristic, outcome and complication including Hirschsprung associated enterocolitis (HAEC) were evaluated in this study. Descriptive analysis is done to determine the characteristic of the subject. The relation between age at surgery and length of bowel resection with HAEC were assessed using independent T-test. The

relation of gender with HAEC was determined using chi-square test.

RESULTS

A total 77 patients include in this study with mean age at surgery was 13.01 months range between 0.3 to 96 months. The majority of patients is male with 43 cases (55.8%) and female is 34 cases (44.2%). The mean of bowel resection was 18.64 cm range from 7-25 cm. The number of HAEC in this study was 42 cases (54.5%). The details of patient characteristic showed in Table 1.

Table 1: Patients characteristic.

Characteristics	TERPT (n=77)
Mean age at surgery in month (range)	13.01 (0.3 - 96)
Gender	N (%)
Male	43 (55.8)
Female	34 (44.2)
Mean length of resection in cm (range)	18.64 (7 - 25)
HAEC	N (%)
Positive	42 (54.5)
Negative	35 (45.5)

The statistical analysis using independent t-test in Table 2 showed significant relation of age at surgery with HAEC incidence (p=0.000) and no significant relation between length of bowel resection with HAEC incidence (p=0.780). Another statistical analysis using chi-square test in table 3 showed there is no significant difference of HAEC incidence between male and female group (p=0.425).

Table 2: Relation between age and length of resection with HAEC.

	HAEC	P value*
Mean age at surgery in month (range)	13.01 (0.3-96)	0.000
Mean length of resection in cm (range)	18.64 (7-25)	0.780

*Statistical analysis using independent t-test.

Table 3: Relation between gender and HAEC.

	HAEC		P value*
	Positive	Negative	
Male	25	18	0.425
Female	17	17	

*Statistical analysis using chi-square test.

DISCUSSION

Traditionally, the definitive treatment for HD has been colostomy followed by several stages of the pull-through procedure. In the low income/ developing countries, both

social and economic burden of HD is becoming a National healthcare burden. The development of new surgical approaches as the treatment of HD is aimed to reduce the cost, length of hospital stay, and morbidity associated with staged pull-through procedures. One stage transanal endorectal pull-through showed successful results as the treatment for HD in developing countries. This procedure provides excellent quality care in one-time hospitalization and prevent the stigma for having a colostomy.⁵

In this study, the age of the subject at surgery range from 0.3–96 months with mean 13.01 months. This finding similar to study by Pratap et al reported the age at surgery ranged from 0.25 to 65 months, with a mean of 16.24 month.⁵ Another study by Hadidi A included 25 babies (<1 month old), 17 infants (1 month-1 year), 19 children (1 year–5 years), and 7 children (5-13 years) of children with HD underwent TERPT. The mean age at surgery was 11 months range from 0.25 to 156 month.⁶

In our study, the majority of patients is male with 43 cases (55.8%) and female is 34 cases (44.2%). A study by Ortiz-Rubio et al reported a literature review TERPT operation on children with HD from MEDLINE and EMBASE between 2005-2012. This study showed the similar result as our study which is the majority of patients is male with 175 cases (64%) from total 297 cases.⁷ A multicenter study by Lu et al in China with total 650 children with HD. This study divided the patients into 2 groups: Neonatal group (n=186) and Non-Neonatal group (n=464). Both groups showed majority of gender was male as reported male/female compassion: neonatal group (120/66) and non-neonatal group (377/87).⁸

In this study the mean of bowel resection was 18.64 cm range from 7-25 cm. A prospective study by Abdul-Ghafoor et al showed the average length of the resected aganglionic segment was (13.9 cm) ranging from (9 - 25 cm).⁹ Another study by Pratap et al showed the mean length of rectosigmoid resection was 30 cm (range, 20-50 cm). The typical resection length in a neonate was 25 and 45 cm in children older than 1 year. The reason to perform a longer resection that extends beyond the dilated and thick-walled bowel is to avoid bowel dysfunction owing to associated hypo- or aganglionosis.⁵

HAEC is a common complication following HD either pre-operative or post-operative. Hirschsprung-associated enterocolitis (HAEC) was first recognized in the late nineteenth century by Hårald Hirschsprung who included it in his hallmark description of congenital megacolon. HAEC is a condition of intestinal inflammation characterized clinically by fever, abdominal distention, diarrhea and sepsis.

Hirschsprung also noted key pathologic findings of HAEC at autopsy; including crypt abscesses, mucosal ulceration, and transmural necrosis. Today HAEC is the leading cause of morbidity and is responsible for half of

the deaths associated with Hirschsprung disease (HD).^{1,10,11}

The number of HAEC in our study relatively high with 42 cases (54.5%). The incidence of preoperative HAEC has decreased over the past several decades from around 44% to about 5% or less in recent years. Postoperative HAEC, which occurs in 2% to 42% of children undergoing definitive procedure, leads to significant morbidity, including increased hospitalization and increased cost.¹² In our study, the mean age at surgery and HAEC showed significant relation ($p=0.000$) with negative correlation ($p=0.000$; $r=-0.569$). This study showed decreased risk for developing HAEC with increased age at surgery. A study by Haricharan et al from Fifty-two children with Hirschsprung disease (median age, 25 days; range, 2 days-16 years). Nineteen (37%) had admissions for HAEC. Proportional hazards regression showed that HAEC admissions decreased by 30% with each doubling of age at diagnosis ($p=0.03$). This study concluded that children diagnosed with Hirschsprung disease at younger ages are at a greater risk for postoperative enterocolitis.¹²

Another study by Lu et al in a total 650 children with HD showed a different number of HAEC incidence after TERPT between neonatal and non-neonatal groups ($p<0.05$). In the neonatal group the number of HAEC is 45 cases (40.2%) compared with non-neonatal groups is 31 cases (10.2%). In this study, the number of other complications also significantly higher in neonatal groups compared with non-neonatal groups: perianal excoriation ($p<0.05$); anastomotic stricture ($p<0.05$) and anastomotic leakage ($p<0.05$).⁸ Another study by Hackam et al¹³ did not show any significant increase in postoperative HAEC with delayed diagnosis.

In our study, the gender ($p=0.425$) and mean length of bowel resection ($p=0.780$) showed no significant relation with HAEC. Although the optimal length of ganglionated bowel resection is not known, a variable length of resection has been suggested in the literature ranging from 2 to 15 cm. The additional resection has been suggested to incorporate colon with abnormal ganglia, with the intent of improving clinical outcomes. A study by Haricharan et al showed no statistically significant difference in the HAEC admissions with length of bowel resection when the group with 5 cm or less ganglionated bowel resection was compared to the group with greater than 5 cm resection ($p=0.52$).¹²

CONCLUSION

The outcome of one stage transanal endorectal pull-through showed the excellent result as the treatment for HD patients in developing countries. The number of costs, hospital stay and complications significantly decreased by performing one stage TERPT compares to the staged procedure.

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