

# Early Experience With Single-Stage Transanal Endorectal Pull Through For Rectosigmoid Hirschsprung's Disease

Shrestha MK<sup>1</sup>, Sherchan M<sup>2</sup>, Dhoubhadel BG<sup>3</sup>, Basnet RB<sup>4</sup>

## Abstract

**Introduction:** Recent trends in surgery for Hirschsprung's disease (HD) have been towards earlier operation and fewer surgical stages. A single-stage transanal endorectal pull through (TEPT) is the latest development and minimally invasive technique for HD. This observational prospective study was designed to evaluate the safety and efficacy of single-stage TEPT technique in the management of rectosigmoid HD. **Materials and Methods:** Infants and children with biopsy-proved aganglionosis and barium enema (BE) documented rectosigmoid HD were included for single stage TEPT. Twenty children aged 22 days to 7 years (17 boys and 3 girls) underwent one stage TEPT over 5 year period. Median follow up was 18 months (range 6-36 months). These patients were evaluated with regard to age, sex, intraoperative details, postoperative functional outcome and complications and need for secondary surgical procedure. **Results:** The operating time was 105-180 minutes (mean 120 minutes). The length of resected bowel was 18-30 cm (mean 25 cm). Intraoperative blood loss was 5-40 ml, less in infants compared to older children. Blood transfusion was needed in none out of 12 infants and 2 out of 8 older children. Transient perianal excoriation occurred in 16 patients. Postoperative enterocolitis occurred in one patient. One patient developed constipation with encopresis following anastomotic stricture. None had adhesive obstruction and prolapse of pulled through colon, one required colostomy for anastomotic leakage and peritonitis while another had retained aganglionic segment and re-do pull through was required. **Conclusion:** Single-stage TEPT can be performed successfully in all age of children. The technique is safe, easily learned and has good clinical results.

**Key words:** Hirschsprung's disease, neonatal intestinal obstruction, single-stage operation, transanal endorectal pull-through

<sup>1</sup>Dr. Manoj Krishna Shrestha, MBBS, MS, Consultant Paediatric Surgeon, Kathmandu Model Hospital, Kathmandu, Nepal, <sup>2</sup>Dr. Mamata Sherchan, MBBS, Medical officer, Siddhartha Woman and Children Hospital, Butwal, Rupandehi, Nepal, <sup>3</sup>Dr. Bhim Gopal Dhoubhadel, MBBS, Medical Officer, Siddhartha Woman and Children Hospital, Butwal, Rupandehi, Nepal, <sup>4</sup>Dr. Ranga Bahadur Basnet, MBBS, MD (Path), Senior Consultant Pathologist, Kathmandu Model Hospital, Kathmandu, Nepal.

## Address for correspondence:

Dr. Manoj Krishna Shrestha  
Consultant Paediatric Surgeon  
Kathmandu Model Hospital  
Kathmandu, Nepal  
E-mail: shresthamanoj@hotmail.com  
Tel: +977-9849982899

## How to cite

Shrestha MK, Sherchan M, Dhaobhadel BG, Basnet RB. Early Experience With Single-Stage Transanal Endorectal Pull Through For Rectosigmoid Hirschsprung's Disease. *J Nepal Paediatr Soc* 2014;34(3):188-194.

doi: <http://dx.doi.org/10.3126/jnps.v34i3.11617>

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## Introduction

Hirschsprung's disease (HD) is congenital aganglionosis of distal intestine. It is the commonest cause of intestinal obstruction in neonates with an incidence of 1 in 4000-5000 newborns. Rectosigmoid HD comprises 75-80% of all HD<sup>1</sup>.

The diagnosis of HD is based on clinical presentation, radiological findings, anorectal manometry and rectal biopsy<sup>2</sup>. Delayed passage of meconium, features of intestinal obstruction in neonatal period, constipation/obstipation in exclusively breast-fed babies warrant HD. Barium

enema (BE) finding of radiological transition zone or rectosigmoid index  $<1$  supports diagnosis of HD<sup>3</sup>. The diagnosis is confirmed by rectal biopsy<sup>2</sup>.

The principle of treatment of HD is resection of aganglionic segment of the gut, pull-through of ganglionic proximal gut and its anastomosis with the anus. Staged-operations i.e., colostomy followed by any one of the abdominal pull-through operations and colostomy closure, have been standard treatment. None of the pull through operations is devoid of complications. There is risk of damage to sacral nerves responsible for fecal and urinary incontinence as well as sexual dysfunction with Swenson's procedure, occurrence of fecaloma in the retained aganglionic segment in Duhamel procedure and high incidence of constipation with Soave procedure<sup>4</sup>. Multistage procedures involve high morbidity and mortality of the patient and prolonged psychological stress of parents. Hence there has been a recent trend towards minimally invasive single-stage primary pull-through for HD<sup>5</sup>.

Dela Torre-Mondragon and Ortega-Salgado in 1998 first reported that pull-through operation can be performed transanally<sup>6</sup>. Many reports have been published confirming safety and feasibility of transanal endorectal pull-through (TEPT)<sup>7,8</sup>. TEPT represents the latest development in the concept of minimally invasive surgery for short segment HD. Laparoscopy or mini-laparotomy can be incorporated in case of long segment HD<sup>9,10</sup>.

The aim of this study is to evaluate the efficacy and safety of single-stage TEPT for rectosigmoid HD in different pediatric age group in our setup.

## Materials and Methods

Over a 5 year-period (July 2008 - July 2013), all the children who were clinically suspected HD underwent BE and punch rectal biopsy to confirm HD. Twenty children aged 22 days to 7 years (17 boys and 3 girls) with biopsy proved HD and BE documented short segment HD were included in the study. Children who presented with colostomy, who needed laparotomy and colostomy, who could not be satisfactorily decompressed by rectal irrigation and needed colostomy and who have long segment disease in BE were excluded from the study.

All the patients were approached as per standard treatment protocol<sup>2</sup> shown in Figure 1 (Fig 1). All clinically suspected patients underwent plain X-ray abdomen in erect posture to rule out pneumoperitoneum. Those who had pneumoperitoneum underwent emergency

laparotomy, multiple biopsies to confirm aganglionic and ganglionic segments and diverting or leveling colostomy. These patients were excluded from the study.

Those patients who had no pneumoperitoneum were subjected to barium enema. We performed barium enema after ruling out pneumoperitoneum at presentation on admission if no rectal manipulation like per rectal digital examination or rectal enema or even suppository had been done. The barium enema was delayed at least for 24 hours if any rectal manipulation was present in order to lessen false positive or false negative findings.

Conservative management consisted of NPO, IV fluid, IV antibiotics and rectal wash out. Rectal wash out with warm normal saline was started after the barium enema was completed. We performed punch rectal biopsy on those patients who improved with the conservative treatment. The biopsy proved and barium enema documented short segment Hirschsprung's disease were included for the study. Those who could not do well with conservative treatment and underwent laparotomy, multiple biopsies and colostomy were excluded from the study.

The patients' demographics, diagnostic work up, operative findings and postoperative findings were recorded.

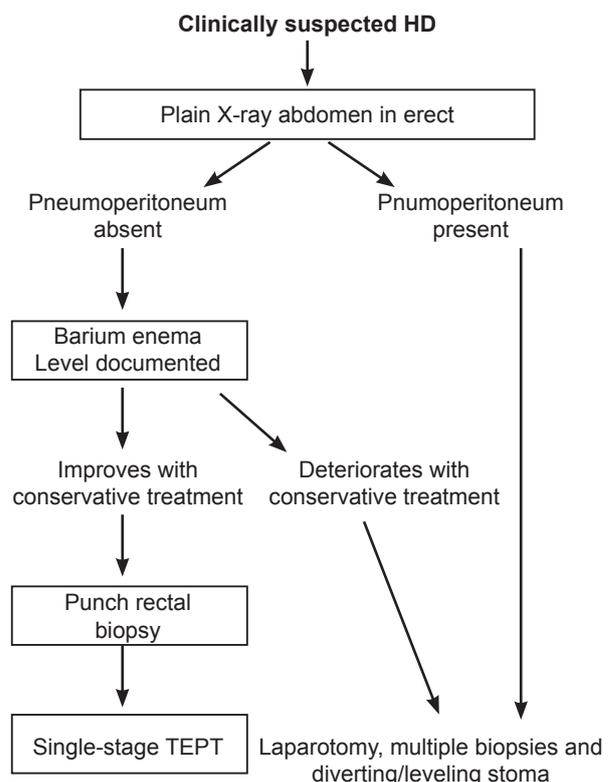


Fig. 1: Treatment protocol for HD.

*Preoperative preparation:* Adequate hydration and intravenous second generation cephalosporin and metronidazole were used as prophylactic antibiotics in all patients. Preoperative bowel preparation consisted of rectal washout with warm normal saline daily. Breast fed babies were allowed breast milk till 6 hours before operation. Older children were kept on low residual diet till 24 hours and clear liquids till 6 hours before operation.

*Surgical techniques:* The operation was done under general anesthesia with endotracheal intubation. A caudal block was used for preemptive and postoperative analgesia. The patient was placed in lithotomy position for older child and suspended lithotomy position for infant and small child (Fig 2). The pelvis was slightly raised with a sand bag/rolled towel under the sacrum. The operation table was slightly inclined to raise the pelvis for better visualization of the anal canal. A bladder catheter was not routinely inserted.

The anus was retracted with stay sutures to expose the anal canal (Fig. 3). Submucosal injection of saline or air was used to ease the initial submucosal dissection<sup>11</sup>. A circumferential incision was given on the anal canal mucosa about 5 mm proximal to the dentate line. Multiple stay sutures were used to hold the mucosal layer as it is dissected circumferentially. Though initial mucosal dissection was difficult and slow, it became easier and faster once the correct submucosal plane was found. The perforating mucosal vessels were cauterized. The submucosal dissection was continued proximally with blunt dissection by traction on the mucosal tube and countertraction on the muscle cuff and cauterization of perforating vessels. After approximately 10-15 cm mucosal tube dissection, there was sudden give way when peritoneal reflection is reached (Fig. 4). Then the muscle of the rectum was incised circumferentially. The muscle cuff was held by two hemostats near midline posteriorly. With the help of a right angle forceps placed posterior to the muscle cuff, it was divided in the midline posteriorly up to the dentate line to prevent constipation. The muscular cuff was excised to shorten the cuff length to about 5 cm<sup>12</sup>. This provided more space for operative manipulation. The rectosigmoid vessels were either cauterized (in infants) or ligated (in older children) and the rectum and sigmoid colon was gradually pulled down through the anus. The pull through of the colon was continued at least 5 cm beyond the transitional zone. The normal colon was dilated, hypertrophied and showed good peristalsis on stimulation (Fig 5). It was cut transversely at this level so that a part of ganglionic colon along with

hypoganglionic and aganglionic segment of colon was excised to avoid retained aganglionic segment. Yes we had no facilities of frozen section biopsy. Ganglionic colon was assessed clinically as it was dilated, hypertrophied and had good peristalsis on stimulation. We preferred excising a few inches more of ganglionic segment than having retained aganglionic segment. We discussed this in discussion section. It was essential that the proximal excised end has good vascularity as well. Several fixation stitches were placed at seromuscular layer near the cut margin and the muscular cuff just proximal to the anal mucosa to prevent retraction of pulled through colon. Colo-anal anastomosis was done using 4-0 polyglactin (vicryl) (Fig. 6). Drains were not used.

#### *Postoperative management*

Feeding was started early on demand, usually 3-6 hours after the operation starting with clear fluid or breast milk and gradually advanced to liquid and soft diet as tolerated. Intravenous fluid was continued till 24 and then gradually tapered. Intravenous antibiotics were continued 72 hours postoperatively followed by oral antibiotics for five days. The patient was discharged (usually at POD 4 or 5) when feeds were well tolerated, flatus and faeces passed regularly, no abdominal distension and/or vomiting and no fever or any abnormal discharge per anus.

Patients were advised to visit for follow up two weeks after operation for rectal examination and routine anastomotic dilatation to prevent anastomotic stricture. The parents were taught to introduce the anal dilator beyond the anastomosis site. The dilation schedule is once daily for one month, on alternate days for one month, twice weekly for one month and once a week for three months.

Follow up visits were arranged at one month, three months, six months and then yearly to look for late postoperative complications like anastomotic stricture, constipation, encopresis, enterocolitis, stooling patterns, retraction or prolapse of pulled through colon, residual aganglionosis, etc. We considered our patients continent clinically if they passed stool regularly at least once daily and they remained dry without fecal soiling, We do not have anorectal manometry or video defecography or other means to assess anorectal function.

## Results

During the five year study period, 20 children underwent one stage TEPT. There were 17 boys and

three girls. The age ranged from 22 days to seven years (mean 18 mo) with 12 infants and 8 older children. Median follow up was 18 months (range 6-36 months).

The operation time ranged from 105 min to 180 min (mean 120 min). It was longer in older children as submucosal dissection was more difficult in older children compared to infants. Intraoperative blood loss was 5-40 ml, more in older children. Blood transfusion was needed in none out of 12 infants and 2 out of 8 older children (>10% of estimated blood volume). There was mucosal perforation in 3 infants during submucosal dissection. This had no effect on infection or outcome of the operation. The length of resected bowel was 18-30 cm (mean 25 cm). The operation events are summarized in Table 1.

Oral feeding consisting of mother's milk or clear fluid was allowed 3 hours postoperatively irrespective of bowel sound. The feeding increased as the patients tolerated. The full feed was achieved by 24- 48 hours. A few episodes of vomiting without abdominal distension were observed in six patients which improved with time without medication. Although presence of bowel sound, passage of flatus or faeces are parameters to start oral feeding, we preferred early enteral feeding as it has several advantages<sup>21</sup>.

Frequency of bowel movement was 2-14 times/day which gradually settled to 1-3 times/day by three months. Transient perianal excoriation occurred in 16 subjects, all improved with medication and decreased frequency of stool with time.

One patient developed constipation with encopresis following anastomotic stricture. He improved with anastomotic dilatation, laxative and toilet training. He was two years old when presented to me, 14 months after the operation. We think this was fecal impaction with overflow incontinence which improved with dilatation of the stricture. Postoperative enterocolitis occurred in one infant who improved with gentle passage of flatus tube, warm NS irrigation and IV antibiotics. One patient required diverting colostomy for anastomotic leakage and peritonitis. One patient had retained aganglionic segment shown by scanty ganglion cells in histopathology report of pulled through colon. He underwent re-do pull-through transanally. None had cuff abscess or prolapse or retraction of pulled through colon. There was no conversion to laparotomy and no mortality. The postoperative complications are summarized in Table 2. Perianal excoriation occurred due to increased frequency of stool complemented by friction on cleaning the stool and superimposed with fungal infection. Perianal excoriation was taken care with application of barrier like Vaseline or oil, antifungal topical cream and it improved as the frequency of stool decreased with time.

**Table 1:** Operation events

Operation events	Infants (< 1 yr) (n=12)	Older children (> 1 yr) (n=8)
Mean operation time (minutes)	113.75	132.5
Mean blood loss (ml)	17.75	23.33
Blood transfusion	0	2
Mucosal perforation	3	0
Mean length of bowel resected (cm)	21.75	30.50

**Table 2:** Postoperative complications

Postoperative complications	n=20 (%)	Infants (< 1 yr)	Older children (> 1 yr)
Perianal excoriation	16 (80.0%)	13	3
Anastomotic stricture	1 (5.0%)	0	1
Enterocolitis	1 (5.0%)	1	0
Anastomotic leakage & peritonitis	1 (5.0%)	0	1
Retained aganglionic segment	1 (5.0%)	0	1
Cuff abscess	none	0	0
Prolapse of pulled through colon	none	0	0
Retraction of colon	none	0	0
Adhesive bowel obstruction	none	0	0



Fig. 2: Patient in suspended lithotomy position

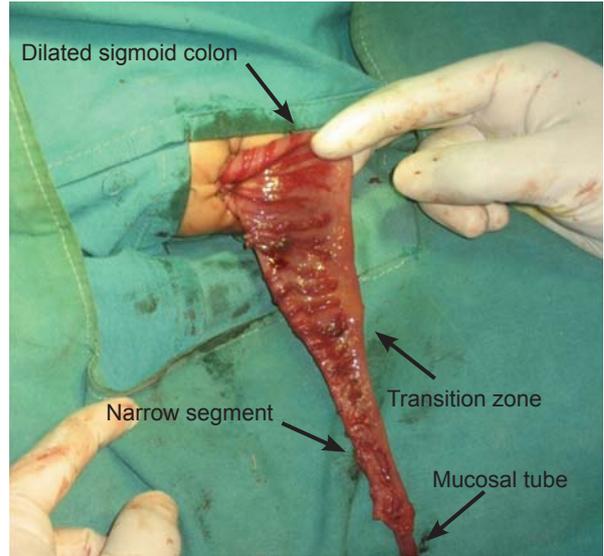


Fig. 5: Dilated sigmoid colon seen during operation



Fig. 3: Showing stay sutures to evert anal mucosa

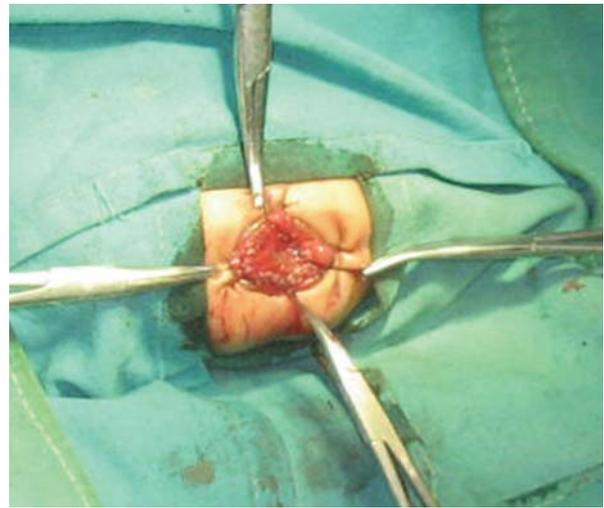


Fig. 6: After completed coloanal anastomosis

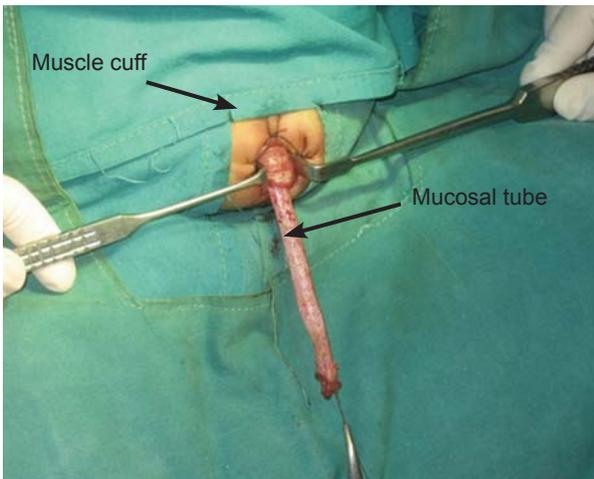


Fig. 4: Site of peritoneal reflection with prolapse of rectum

### Discussion

The diagnosis and treatment of HD have always been a challenge to pediatricians and pediatric surgeons. Increasing awareness of presentation and high index of suspicion can lead to early diagnosis. Biopsy-proved aganglionosis and properly performed BE correlated with the level of aganglionosis are prerequisites for TERPT<sup>13</sup>.

The use of complete transanal pull through for HD was first described by Dela Torre-Mondragon and Ortega-Salgado in 1998<sup>6</sup>. Many reports have been published since then from different parts of the world. The safety, feasibility, reproducibility and less invasiveness of the procedure made it popular in recent years<sup>9,14,15,16,17,18</sup>.

Initial submucosal dissection was slow and difficult. It was facilitated by submucosal injection of air or saline<sup>11</sup>. In our series, we found that the submucosal dissection was easier in neonates and infants compared to older children. The operation time and blood loss were less in neonates and infants. This was probably due to more adhesion resulting from recurrent enterocolitis and bigger vessels in older children. The operative time of our series is comparable with several other studies<sup>7,8,9</sup>.

We found increased frequency of bowel movement (2-14 times/day) after TEPT which gradually settled to 1-3 times/day by 3 months. Transient perianal excoriation occurred in 16 subjects; all improved with medication and decreased frequency of stool with time. Similar finding was reported by other authors<sup>7,19</sup>.

We had anastomotic stricture at the beginning of the series. The dilatation of the stricture was difficult for the parents as they could not accurately pass the dilator through the stricture. So we started routine prophylactic anastomotic dilatation similar to anal dilatation following Pena procedure for ARM to prevent postoperative anastomotic stricture. Routine anastomotic dilatation was easy and parents could perform it correctly. Since then we did not have anastomotic stricture. Routine anastomotic dilatation has been recommended by several other authors<sup>7,18,20</sup>.

Enteral feeding<sup>21</sup> was done and there were a few episodes of vomiting without abdominal distention in six subjects which improved with time without medication. This may be because of the side effects of the medicines we used. We had multiple episodes of vomiting with abdominal distension in two subjects because of enterocolitis in one and anastomotic leakage and peritonitis in the other. They improved with the treatment of the cause.

There was one incidence of retained aganglionic segment. The histopathology of the distal pulled through colon, which was routinely done to confirm the presence of ganglion cells in the pulled through colon, showed scanty ganglion cells. Parents were warned that the child might have the previous symptoms and might need second operation if symptoms recurred. He developed the symptoms after 4 months and we managed by re-do TEPT. Ideally, frozen section biopsy of the distal pulled through colon should be done before colo-anal anastomosis. Since there were no facility of frozen section biopsy in the country (except one institution) it was not done. On the other hand, frozen section biopsy is not foolproof; it does have false positive results<sup>22</sup>. In developing country like ours,

we depend clinically to see the dilated, hypertrophied bowel with peristaltic movement on stimulation. To be on safe side we resect about 5 cm of the clinically ganglionic segment<sup>7</sup>.

There were no complications like cuff abscess, prolapse or retraction of pulled through colon, adhesive bowel obstruction as reported in other series<sup>9,23</sup>.

## Conclusion

Single-stage TEPT is minimally invasive, safe and effective procedure for rectosigmoid HD in different pediatric age group in our setup. Moreover it can be learnt quickly and reproducible.

**Acknowledgement:** Nil

**Funding:** Nil

**Conflict of interest:** None

**Permission from IRB:** Yes

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