



Original Article

IS SINGLE-STAGE PRIMARY TRANSANAL ENDORECTAL PULL THROUGH (TEP) FEASIBLE FOR SHORT SEGMENT HIRSCHSPRUNG'S DISEASE IN NEONATES?- OUR EXPERIENCE IN A TERTIARY HOSPITAL

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Abstract

Background: The surgical management of Hirschsprung's Disease (HD) includes so many procedures. Transanal Endorectal Pull through (TEP) represents the latest development in the concept of the minimally invasive surgery for HD. The purpose of this study was to evaluate the outcome of the procedure in neonates.

Methods: This retrospective study was carried out in a tertiary pediatric hospital during the period from January 2007 to December 2012 (5 years). The study included neonates of both sex, weight more than 2 kg, who were clinically suspected HD, radiologically transition zone at rectosigmoid and midsigmoid region and rectal biopsy proven HD and no evidence of sepsis or enterocolitis. Short segment HD with associated anomalies, and operated cases with less than 6 months' or irregular follow up were excluded.

Results: During study period, single-stage transanal endorectal pull through (TEP) operation was done for short segment HD in 63 neonates, 9 patients were excluded from the study for irregular follow up. The mean operative time, mean blood loss, postoperative hospital stay, follow up period were 113 minutes, 20ml, 6.8 days and 19.6 months respectively. Transverse colostomy was needed in 6 patients for anastomotic leakage, thereafter developed anastomotic stricture, managed with regular anastomotic dilatation.

Conclusion: Advancement in pediatric anesthesia, improvement of pediatric surgical expertise, perioperative management and nursing care has made single-stage primary transanal endorectal pull-through a feasible and safe surgical procedure for the treatment of short segment Hirschsprung's disease in neonate.

Keywords: Single-stage transanal endorectal pull through; short segment Hirschsprung's disease in neonate.

Introduction

Hirschsprung's Disease (HD) is suspected when a newborn fails to pass meconium in the first 48 hours of life, has abdominal distention and bilious vomiting¹. Clinical history, radiological study, anorectal

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manometry and rectal biopsy are the basic diagnostic tools of HD². The surgical management of HD includes multistaged procedures and also single-stage procedures like- laparotomy with single-stage pullthrough³; primary laparoscopic assisted endorectal pull-through⁴ and primary transanal endorectal pull-through (TEP) without colostomy⁵⁻⁸. Techniques may vary but operative principles are same: resection of aganglionic segment, pull-through of the ganglionic proximal gut and anastomosis of the ganglionic gut with the anorectum with or without initial colostomy and subsequent closure depending on single or staged procedure.

All of the procedures for the definitive treatment of HD have more or less some complications⁹. Swenson, have complications like- damage to sacral nerves responsible for fecal and urinary incontinence as well as sexual dysfunction, Duhamel and Soave procedures have occurrence of fecoloma in the retained segment and high incidence of constipation¹⁰. Multistage procedures involve high morbidity and mortality of the patients and prolonged psychological stress of the parents. Hence there has been a recent trend towards minimally invasive single-stage primary pull-through for short segment HD¹¹.

Primary transanal endorectal pullthrough prefers to eliminate the need for laparotomy, therefore risk of intestinal adhesion and obstruction. It offers the advantages of staying away from pelvic structures, preserving internal anal sphincter, blood supply and innervation, thus urinary and fecal continence are less likely to be offended¹². It has advantage of less operating time, intraoperative blood loss, post-operating pain and also gives excellent cosmetic results as well as good parent compliance⁷. It reduces hospital stay and workload. It is more cost effective, with least psychological stress, good parental satisfaction and compliance⁵. Although there is still clear indication for decompressive colostomy in some situations, primary pull-through approach should be the current standard care for patients with classic short segment HD¹¹.

Recently there are so many literatures found on single stage TEP in neonates and infants. The purpose of the study was to evaluate the outcome of single stage transanal endorectal pull-through for short segment Hirschprung's disease in neonates in the context of our country.

Materials and Methods

This retrospective study was carried out in the division of neonatal surgery, Department of pediatric Surgery, Dhaka Shishu (Children) Hospital during the period from January 2007 to December 2012 (5 years). The study included neonates of both sex, weight more than 2kg, who were clinically suspected Hirschprung's disease, radiologically transition zone at rectosigmoid region and rectal biopsy proven HD, no evidence of sepsis or enterocolitis. Patients with short segment HD who had associated anomalies and hugely dilated proximal colon, operated cases with less than 6 months follow up and irregular follow up were excluded.

The colon was prepared before operation as per following protocol: nothing per oral and IV infusion

started the day prior to surgery, mechanical cleansing and colon decompression by rectal irrigation twice daily with warm normal saline for 1 to 3 days. Intravenous antibiotics (Ceftazidime + Metronidazole) were given immediately before surgery.

Surgical Technique

After the induction of general endotracheal anesthesia and the placement of intravenous lines, the patients were given combination of one of the third generation cephalosporins and metronidazole intravenously. Rectal irrigation was performed with a dilute solution of povidine iodine. The patient was then placed supine with the pelvis elevated at the end of the operating table with the lower limbs attached to an inverted U-shaped bar. The abdomen and perineum were prepared in the standard fashion. A bladder catheter inserted. The anal canal was exposed with Loan-Star anal retractor. Submucosal injection of epinephrine or saline was not routinely used. A circumferential row of 4-0 silk stay sutures was inserted approximately 0.5 to 1 cm above the dentate line. The rectal mucosa was incised just distal to the traction sutures and lifted circumferentially using fine diathermy needle to develop the submucosal plane. Once the submucosal plane was established, the dissection was easily continued proximally using blunt dissection and cauterization of submucosal infiltrating vessels. The traction on mucosal tube facilitated proximal extension of mucosal dissection until the level proximal to peritoneal reflection (approximately 10 to 15 cm above the dentate line). Four stay sutures were inserted to control the full-thickness sigmoid colon. Mobilization of the colon was continued as proximal to the grossly obvious or histologically confirmed transition zone as possible by dividing the rectosigmoid vessels after cauterizing them. Ligatures were rarely needed in neonates. The long seromuscular cuff was inverted outside the anus, posteriorly slitted longitudinally and repositioned to its normal position. After resection of the aganglionic segment, the normally innervated bowel was pulled through the muscular cuff and anastomosed to the remaining mucosa above the dentate line using 4-0 slowly absorbable suture material. Feeding was allowed at first on the second postoperative day. The patients were discharged 3-5 days after surgery in the absence of any complications. First rectal digital examination was performed after 14 days. If the colorectal anastomosis was adequate we demonstrated the dilatation procedure to the parents

with the metallic anal dilator no-3 (Hegar's dilator no-11/12) size that snugly passed through the colorectal anastomosis and advised anastomotic dilatation by anal dilator by the parents at home according to dilatation schedule for anorectal malformation by Pena¹⁴. The parents were advised to bring their baby for followup at 1 month interval for 3 to 4 months. If the coloanal anastomosis was too tight for digital dilatation during the follow up period, we examined the patient under general anesthesia. In case of anastomotic stricture, dilatation under general anaesthesia was advised along with twice daily home dilatation by metallic dilator number-3. In the absence of any anastomotic stricture we advised home dilatation by anal dilator and more frequent follow up visits.

All the data were collected from operation room, departmental and hospital record books for further analysis, which included age, sex, clinical presentations and investigations, operative details, early post operatives findings, post operative complications, functional outcome, any need for a secondary surgery and follow up time.

Results

During the study period of five years 63 neonates were included for clinical study following the selection criteria. 9 cases were excluded from the study for irregular follow up. The mean age of the study population in this series was 16.4 days (range; 2 - 28 days). There were 35 (65%) male and 19 (35%) female patients. On contrast enema transitional zone was found in Rectum in 9 (17%), Recto sigmoid colon in 33 (61%) and Sigmoid colon in 12 (22%) patients. Operative variables (operative time, blood loss, length of resected bowel), post operative hospital stay and

postoperative bowel movement are given in Table-I and 2. Postoperative complications and their management are given in Table-III. Sixty three neonates were operated within the study period. But nine of them had irregular follow up and excluded. Regular post operative mean follow-up was 19.6 months (18-36 months).

None of the patients developed disturbances of micturition. 2 patients presented with features of post-operative constipation and post pull-through enterocolitis.

Parents of all patients were inquired during follow up visits. Most of them expressed satisfaction with the operative outcome in terms of expected cosmesis, hospitalization, and absence of persistent symptomatic postoperative complications.

Table-I
Operative variables (n=54)

	Range	Mean
Length of resected bowel (cm)	9 to 23	14.32
Length of rectal muscle cuff (cm)	3 to 6	4.4
Operation Time (min)	90 to 130	113
Blood Loss (ml)	10 to 27	22.3
Post operative Hospital Stay (day)	4 to 14	6.8

Table-II
Postoperative bowel movement (n=54)

Time (hours)	No of pts	Percentage
12-24	30	56
24-48	18	33
48-72	6	11

Table-III
Postoperative complications (n= 54)

Complications	No of pts.	Management
Anastomotic leakage followed by development of anastomotic stricture	6 (11%)	Managed initially with conservative treatment followed by transverse colostomy. For anastomotic stricture dilatation under GA for 1 st time and subsequently regularly at home.
Perianal excoriation	28 (52%)	Local application of Zinc Oxide Paste.
Enterocolitis and constipation	2 (4%)	Managed initially conservatively and then by posterior myectomy.

Discussion:

Hirschsprung's disease is a common surgical problem in pediatric patients. In majority of them, the diseased segment extends up to the recto-sigmoid junction. The treatment of Hirschsprung's disease is to resect the aganglionic segment of the rectum and colon, pull-through normally innervated colon and anastomose this bowel at the anorectal region preserving the sphincter muscle. The disease can be managed by traditional multi staged procedures with colostomy and single-stage procedures without colostomy. Single-stage TEP procedure is the concept of the minimally invasive surgery and the latest developmental new era¹².

Transanal pull-through is relatively simple and advantageous in newborns, in whom the colon is loosely fixed to the retroperitoneum allowing longer segments of the colon to be resected through the anus in contrast to the more laborious dissection in older patients¹³. In our study the mean length of bowel resected was 14.32 cm, which is comparable to other studies¹⁴⁻¹⁶. The operating time and intraoperative blood loss are also lower in neonates as compared to older children due to the presence of less adherent mucosa, lesser fat laden mesentery, less dilated colon above the aganglionic segment, easily controllable blood vessels and less episodes of enterocolitis¹⁶.

Total 54 neonates were included in this study of whom, mean age was 16.4 days (range from 2 to 28 days). Among them 35 were male and 19 were female with M:F ratio of 1.8:1. Near similar male preponderance was found in other studies^{12,17}.

The mean length of resected aganglionic segment of bowel was 14.32 cm (range 9 to 23 cm). Studies from Egypt⁸ and China¹⁸ stated, the length of resected segment was about 12.5 to 45 cm.

The long cuff is often blamed for post-operative obstructive symptoms, constipation and enterocolitis¹⁹. The length of rectal muscle cuff (sleeve) in this study was 4.4 cm (range 3 to 6 cm). In others studies^{7,12,18} the retained cuff length was 3-6 cm, which was responsible for constipation. We have two patients with such complications, initially managed conservatively then with posterior myectomy.

In our study, the mean operative time for pull-through including frozen section biopsy was 113 minutes (range 90 to 130 minutes). Another study in Dhaka Shishu (Children) Hospital²⁰ showed that the mean

operative time for pull-through was 141.56 minutes (range 125 to 175 minutes). Some other studies showed mean operative time ranging from 96 minutes to 186 minutes⁵⁻⁸. Relatively longer durations were probably due to waiting for the results of the frozen section biopsies and time deferred in different expertise.

In our study, postoperative fever occurred in 32 patients. Among them, 26 (48%) patients had low grade fever lasted for 2-4 days. Fever in early post operative period developed probably due to operative trauma, intravenous infusion and chest infection. Six patients who had anastomotic leakage developed high fever and persisted for 7 days.

In most of the patients, bowel moved between 12-24 hours after operation. In the study, frequent bowel movement of loose type was seen in the immediate postoperative period in most of the cases, 10 to 15 times a day with perianal excoriation which was reduced to 4-6 times a day within 3 months. One of the major issues in the transanal approach is the significant stretching of the anal sphincters during surgery with its potential impact on continence in later life. However, this is only transient and bowel movements became normal in majority of cases within a period of 2 weeks to 3 months²¹. In other studies^{9,22}, the frequency of stool immediately after the operation was 5 to 8 bowel movements per day, which declined to a mean 2-3 times/day after 3 months. Transient increased frequency of bowel motions does occur probably due to stretch effect on sphincters. This transient effect becomes normal in the majority of cases within 3 months⁸.

In this study, 28 patients developed mild to severe perianal excoriation in the early postoperative days and resolved within 3 months (Table-3). Among these patients, 5 had severe perianal excoriation for which hospital stay had been prolonged (13 days). Remaining 23 patients had minimal perianal excoriation on discharge. This was due to frequent loose bowel movement. Subsequently with reduced frequency of bowel movement and treatment, excoriation was resolved within 3 months. Study in United States²² and England²³ reported perianal excoriation was common (42% and 49%) but generally resolved within 3 months.

In an American study³, the anastomotic leakage was 8%. In this study, 6 patients (11%) developed anastomotic leakage, which were managed initially

by conservative treatment and later on doing transverse colostomy. These six patients developed postoperative anastomotic stenosis and resolved by anastomotic dilatation under general anesthesia for the first time and then daily home dilatation by mother; colostomies were closed after 6-8 weeks. In Missouri³ and Liverpool⁹ studies, 20% and 2.49% patients respectively had additional surgical procedures for postoperative complications after TEP respectively. The authors did not mention the indications for additional surgery for postoperative complications after TEP.

In our study, no patient developed urinary disturbances or fecal incontinence or soiling, due to rectal muscle cuff. None of them developed adhesive intestinal obstruction after pull-through in our series. A study in Egypt⁸ adhesive intestinal obstruction occurred in 0.7% patients.

Two patients (3.7%) had constipation with enterocolitis. A study in Egypt, postoperative enterocolitis occurred in 17.8% cases. Enterocolitis has been considered one of the common problems in patients with HD both before and after definitive treatment⁸.

There was no death in our series. This may be attributed to proper selection of patients for TEP, skilled personnels, proper management and small number of patients.

In our study, most parents were satisfied with the pull-through procedure, in terms of bowel pattern normalization, absence of persistent postoperative complications and cosmetic appearance as per their own expectation.

Conclusion

With the development and advancement of neonatal anaesthesia, surgical expertise, pre and perioperative nursing care, one stage primary transanal endorectal pull-through for short segment HD is now feasible in neonates. It preserves all of the advantages of minimally invasive surgery. TEP is a better option in terms of economy, comfort, operative success, postoperative morbidity, cosmetic appearance and parents satisfaction.

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