Original Article

Outcome Analysis of Single-Stage Transanal Endorectal Pull Through in Selected Patients with Hirschsprung Disease

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Abstract

Background: Hirschsprung disease is a notable cause of neonatal intestinal obstruction and constipation in older children. Transanal endorectal pull through (TEPT) is a newer technique of definitive management as against staged procedures. The aim of our study is to evaluate the feasibility and outcome of the procedure in selected children with Hirschsprung disease managed by this technique with review of the literature. **Materials and Methods:** Medical records of 12 children who underwent single-stage TEPT in a tertiary care centre over a period of 3 years from 2015 to 2018 were reviewed and retrospectively analysed on the basis of age, investigations, intraoperative parameters, complications, functional outcome and hospital stay. **Results:** The median age at surgery was 9 months. Nine patients were boys. The median weight of patients was 7.5 kg. The transition zone was observed at the level of the rectosigmoid in eight patients (66.6%) and sigmoid colon in four patients (33.3%). The mean length of muscle cuff was 3 cm, the mean length of resected bowel was 25 cm, the median operative time was 105 min and the mean hospital stay was 8 days. Perianal excoriation (n = 2) and enterocolitis (n = 1) were complications encountered postoperatively; however, no patient had cuff abscess, anastomotic leak or stricture. Stool frequency initially at 2 weeks was average of six to ten times a day, which gradually reduced to two to three times a day by 3 months postoperatively. None of the patients had faecal soiling or constipation on follow-up. **Conclusion:** Single-stage transanal endorectal pull through is an effective technique in the management of Hirschsprung disease with minimal complications.

Keywords: Colorectal, hirschsprung disease, paediatric, transanal endorectal pull through

Introduction

Hirschsprung disease is a frequent cause of intestinal obstruction in neonates and constipation in older children. Various surgical techniques have evolved over time for the definitive management of Hirschsprung disease, single-stage Transanal endorectal pull through (TEPT) being one of the recent techniques. The study is aimed at evaluating the feasibility and outcome of the procedure. We present the outcome analysis in 12 children with Hirschsprung disease managed by single-stage TEPT over a period of 3 years from 2015 to 2018 with regard to technique, functional outcome and complications. They were followed up for a period of 2 years.

Materials and Methods

Medical records of 12 children who underwent one-stage transanal endorectal pull through at a tertiary care centre from 2015 to 2018 were reviewed and retrospectively analysed on the basis of age, investigations, operative parameters, complications, functional outcome and duration of hospital stay. Patients with clinical suspicion of Hirschsprung disease were put on rectal washouts and investigated with a barium enema to look for the presence and level of radiological transition zone (TZ). Those with the presence of TZ underwent full-thickness rectal biopsy (FTRB). Only those patients who were deflating well on regular washouts; the patients whose barium enema [Figure 1] suggested TZ in rectum, rectosigmoid or sigmoid colon and with histopathologically confirmed aganglionosis at FTRB were included in the study. The patients

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Figure 1: Barium enema showing level of transition zone (original)

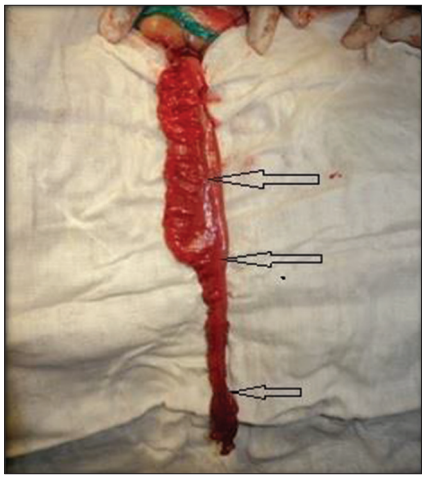

Figure 2: Intraoperative-aganglionic rectal segment, transition zone and dilated sigmoid colon (original)

who presented late with a dilated sigmoid colon; those who were non-compliant with rectal washouts and with radiologic TZ proximal to sigmoid colon were excluded from the study.

Preoperatively, the patients were kept nil orally 24 h before surgery to prevent intraoperative and early post-operative wound contamination. Double catheter rectal washouts with warm saline were administered for bowel preparation. Prophylactic intravenous broad-spectrum antibiotics were administered 1 h before surgery to cover gram-negative bacilli and colonic anaerobes.

Operative procedure

The patients were placed in lithotomy position with a pelvic tilt; the bladder was catheterized and the anal canal was exposed by the use of stay sutures at the anal verge. Mucosal stay sutures were placed 1.5 cm proximal from the dentate line. A circumferential incision was made 1 cm proximal to the dentate line followed by rectal mucosal dissection in the submucosal plane for 3 cm. The mucosa was stripped from the underlying muscle, initially using fine electrocautery and subsequently using blunt dissection. After the mucosal dissection was completed, the rectal muscle was incised circumferentially. Dissection was then continued full thickness by dividing fibrovascular bands, and proximal bowel was telescoped through the muscular sleeve [Figure 2]. The vessels were divided just as they entered the bowel wall, to avoid injury to pelvic nerves, as well as the prostate gland or vagina. The principle of surgery is to resect aganglionated bowel segment; pull through and anastomosis of ganglionated bowel segment. Multiple full-thickness biopsies were sent for the frozen section to define the level of TZ and ganglion cells in proximal dilated bowel. Once the normally innervated bowel was reached, the bowel was divided and aganglionic segment resected. The length of muscular cuff was measured by thin, sterile surgical ruler. The muscle cuff was split at 6 o'clock position. The ganglionic part of the colon was fixed within muscle cuff from below. Then, the coloanal anastomosis was completed.

Postoperatively, the patients were kept nil by mouth with intravenous antibiotics for 5 days to prevent possible stool contamination and resultant anastomotic complications. Perianal skin hygiene was strictly maintained, and petroleum jelly was applied locally as barrier cream. The patients were kept on regular follow-up for 2 years, and parents were asked to note the stooling pattern. No patient was lost to follow-up.

The data obtained from the patients' medical records were analysed with respect to age at surgery, investigations, operative parameters, complications, functional outcome, duration of hospital stay and follow-up.

Results

In our study, the median age at the time of surgery was 9 months (range: 6 months–3 years). Nine boys and three girls underwent surgery. The median weight at surgery was 7.5 kg (range: 6.4–10 kg). TZ was at rectosigmoid in eight patients (66.6%) and the sigmoid colon in four patients (33.3%) as seen on barium enema. Intraoperatively, the mean length of muscle cuff was 3 cm, and the average length of resected bowel was 25 cm. The median operative time required for the procedure was 105 min. The patients stayed in the hospital for an average of 8 days. None of the patients required laparoscopy or laparotomy.

In the post-operative period, two children (16.6%) had perianal excoriation and one child (8.3%) had enterocolitis, which responded to medical management. The patients were kept on follow-up for 2 years. No patient had a cuff abscess, anastomotic leak or stricture. Stool frequency initially at 2 weeks was six to ten times a day and gradually reduced to two to three times a day by 3 months post-operatively. No patient had faecal soiling or constipation.

Discussion

Treatment options for Hirschsprung disease have evolved from a staged procedure-colostomy followed by definitive surgery to a single-stage procedure. The latter has been observed to be comparable or even better than the staged procedure.[1]

TEPT when done as a one-stage procedure avoids multiple anaesthesia exposures, exempts the morbidity of stoma and reduces the cost.

TEPT represents a natural evolution from the laparoscopic procedure.[2] The minimal access approach for Hirschsprung disease was first described by Georgeson *et al*. in the early 1990s wherein the procedure consisted of a laparoscopic biopsy to identify the TZ, laparoscopic mobilization of the rectum below peritoneal reflection and a short endorectal mucosal dissection from below.[3] The initial series of children with Hirschsprung disease was published by de La Torre-Mondragon and Ortega-Salgado and Langer *et al*. in the late 1990s.[4,5] Many studies published later have proved the safety, efficacy, cost-effectiveness and good functional outcome of the procedure.

Single-stage TEPT has a precise indication; hence, case selection is very important – cases with TZ involving rectum and sigmoid colon are most suitable for this procedure, parents should be compliant with rectal washes and colon should be effectively decompressed with washes.

In most cases, TEPT is performed in infancy. There are series, in which TEPT is performed in neonatal age group.[6] In our study, the median age at the time of surgery was 9 months (6 months–3 years). TEPT can be performed successfully in all ages of children with good results, avoiding abdominal exploration.[7] Almost all studies regarding TEPT showed male preponderance over females. This might be due to the higher incidence of Hirschsprung disease (especially short segment Hirschsprung disease) in males as compared to females (M:F = 4:1). Similar findings regarding male preponderance were noted in our study with nine boys and three girls.

Although contrast study is commonly used to identify the level of TZ, it is not accurate in locating the pathological transition zone. In 12% of cases, pathologic TZ is different from the radiological TZ.[8] The accuracy of contrast enema in identifying the level of TZ in older children may be improved by discontinuing the rectal irrigations for 1–3 days before the study. By discontinuation of the washes, adequate time is offered for the proximal bowel to distend and demarcate the TZ on contrast enema.

Tannuri *et al*. in their series on TEPT have reported a refinement in technique by not giving preoperative bowel preparation.[9] However, we have followed the technique of bowel preparation in our study, as per the classical technique to avoid wound contamination and dehiscence. The mucosal incision above the dentate line depends on the size of the child, but it is crucial that the incision is high enough above the dentate line so that the transitional epithelium is not damaged.[10] This is important to prevent the loss of sensation, which may predispose the child to long-term problems with incontinence. Langer *et al*. state that it ranges from 0.5 to 1.0 cm above the dentate line in a new born and 1.0–2.0 cm above the dentate line in an older child.[11] The present technique involves proceeding with a short mucosal dissection for 1.0–3.0 cm and then incising the rectal wall circumferentially. With a very short cuff, the muscle does not need to be incised in most cases. Some surgeons have eliminated the mucosal dissection entirely and performed a transanal Swenson procedure.[12] The advantage of leaving a short cuff or no cuff is the avoidance of a constricting ring or residual aganglionic bowel, with a lower risk of obstruction and enterocolitis.[13] The disadvantage is that dissection on outside of the rectum deep in the pelvis may increase the risk of injury to pelvic nerves and vessels, prostate gland, urethra or vagina. Initial descriptions of TEPT involved a long rectal cuff, but it may either constrict the pulled through bowel or roll down into a ring during the pull through; hence, a shorter cuff is preferred now.[14]

The length of resected bowel depends on the length of aganglionic bowel segment. Teeraratkul, Isa *et al*. and Pratap *et al*. reported the length of resected bowel to be 9–25 cm, 18.64 cm and 30 cm, respectively, which is comparable to our study.[15-17] The operative time and as a result, the overall anaesthesia time can range from 95 min in some studies to about 180 min in some other studies.[15,17] The average operative time in our study was 105 min. Operative time included the process and reporting of the frozen section to confirm the presence of ganglion cells.

At least 50% of children develop perianal dermatitis because of frequent bowel movements and liquid discharge during the initial months after a transanal pull-through operation. It is important to prevent this as much as possible by immediate application of barrier creams and in some cases antidiarrheal medication. Increased stool frequency and perianal excoriation both are known to settle down within several weeks to months post-operatively.[2]

The most important and dangerous complication after a pull-through procedure is enterocolitis.[18] One patient developed enterocolitis 1-month post-surgery in our study. It was managed with intravenous antibiotics and adequate hydration. Many preventive measures have been described including routine post-operative irrigations or rectal stimulation, the use of intravenous antibiotics such as vancomycin and metronidazole and also administering probiotics.[19,20] Menezes *et al*. have reported the incidence of obstructive symptoms to be 8%–30% after TEPT.[21] However, we did not encounter obstruction/constipation in any of our operated children. These obstructive symptoms can be taken care of by bowel management program, after stricture and residual aganglionosis is ruled out.

There are some intraoperative difficulties encountered during TEPT-narrow field of vision, retraction of vessels if adequate care is not taken, stretching of anal sphincters, TZ seen in pre-operative barium enema may be located at a higher level intraoperatively, thus making it difficult to reach the ganglionic colon.[8] However, the advantages of TEPT-minimal access approach, negligible risk of intra-abdominal adhesions, good

cosmesis as there is no abdominal scar, bowel not opened intra-abdominally or intraperitoneally, well preserved pelvic structures, sphincters, local blood supply and innervation, thus no effect on faecal and urinary continence. Furthermore, there is a significant decrease in need of analgesics in immediate post-operative period and a decreased total hospital stay and better cosmetic outcome.[22-24]

There are a few limitations of this study – retrospective analysis in a small cohort of patients. We acknowledge that large population-based studies/randomized control trials would be needed for better analysis.

Conclusion

Single-stage transanal endorectal pull through for the management of rectosigmoid and sigmoid Hirschsprung disease is feasible and may be preferred in carefully selected patients. The safety and cost-effectiveness of this procedure is of special interest for developing countries. The functional outcome after the procedure is highly satisfactory.

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Conflicts of interest

There are no conflicts of interest.

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