Original Article

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Transanal Endorectal Pull-through for Hirschsprung's Disease During the First Month of Life

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Background/Purpose*: Hirschsprung's disease (HD) is a common cause of bowel obstruction in the newborn period. One-stage surgery for HD is well established and the results are comparable or better than multistage surgery. The aim of this study was to test the feasibility and safety of transanal endorectal pull-through (TEPT) for management of HD during the neonatal period.*

Materials & Methods*: twenty eight neonates having HD were treated with TEPT at pediatric surgery unit, Mansoura University Children's Hospital (MUCH) during the period from May 2007 to Jun 2009. Six cases were in need for concomitant laparotomy due to long segment disease. Endorectal mucosectomy was started one cm. above dentate line and continued till the peritoneal reflection. The affected bowel was resected and colo-anal anastomosis was performed with 4/0 absorbable sutures.*

Results*: The mean operative time was 90±18 minutes. Blood transfusion was not needed. Oral feeding started 24-48 hours postoperatively and the mean hospital stay was 3-5 days. The commonest postoperative complication was perianal excoriations (64.3%), anastomotic leak occurred in one case. Two cases were in need for repeated dilatations while 4 cases presented with postoperative enterocolitis (EC).*

Conclusion*: TEPT during the neonatal period is easy, bloodless, without visible scar and with short intraoperative time and postoperative hospital stay.*

Index Word*: Hirschsprung's disease, transanal endorectal pull-through, neonates.*

INTRODUCTION

Hirschsprung's disease (HD) is a common cause of bowel obstruction in the newborn period. The time-honored approach to therapy was to perform a preliminary colostomy in the normally innervated bowel in the neonatal period and subsequent definitive pull-through at a later date (at 6 to 15 months of age). More recently, pediatric surgeons are performing primary pull-through procedures without a colostomy in the neonatal period [1].

One-stage surgery for HD is well established and the results are comparable or better than multistage surgery [2,3,4]. However, it is still universally accepted that a preliminary colostomy may be indicated for children with sever enterocolitis (EC), malnutrition, perforation or massive dilatation of proximal bowel [5].

Primary endorectal pull-through in the newborn period was first described by so et al in 1980 (6) with 81% of the patients were totally continent after 18-year follow-up [1].

Total transanal surgery for HD was first reported by

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De La Torrre and Ortega in 1998 [7]. Many reports have confirmed the feasibility of this approach [8-11]. It represents the latest development in the minimally invasive surgery for HD [7,8,11-16].

TEPT is safely be performed during the neonatal period and early infancy and it has the advantage of shorter hospital stay (and therefore less costs), avoid colostomy with its associated complications with satisfactory postoperative results especially for continence [17].

The aim of this study was to test the feasibility and safety of transanal endorectal pull-through for management of HD during the neonatal period.

PATIENTS AND METHODS

Twenty eight consecutive patients having HD were admitted at pediatric surgery unite, Mansoura University Children's Hospital, Mansoura, Egypt, during the period from May 2007 to Jun 2009. All the patients were in the neonatal period with age ranged from 5 days to 30 days at operation. They were subjected to totally transanal endorectal pull-through (TEPT) except for 6 neonates that were in need for concomitant minilaparotomy to mobilize the colon due to long segment disease (transitional zone was not apparent, or present beyond the rectosigmoid junction).

The case files were evaluated for age at operation, associated congenital anomalies, operative time, intraoperative blood loss and blood transfusion, time to resume oral feeding, hospital stay, postoperative complications, and postoperative continence state.

The median follow-up period was 8 months.

The diagnosis was confirmed by barium enema and partial thickness rectal biopsy for histopathological examination for all cases. The cases presented with enterocolitis (n=7), were treated first with rectal washout and third generation cephalosporins.

Preoperative bowel preparation started 48 hours before surgery with rectal irrigation using normal saline every 12 hours.

The newborn was kept on non-residue diet 24 hours before surgery.

Third generation cephalosporin was administered at induction of anesthesia and continued for 3 days after operation.

Surgical procedure:

The infant is placed in prone position for 15 cases and supine position for 13 cases at the end of the operating table. The prone position provided good exposure to the anus with easier control of the vascular pedicle, but when the transitional zone was not apparent on barium enema (7 cases) or barium enema revealed transition zone proximal to recto-sigmoid junction (6 cases), supine position was adopted for the possibility to do concomitant laparotomy for more extensive mobilization of the colon.

A urinary catheter was placed in the urinary bladder. The anal canal was exposed with 4 stay sutures. Normal saline solution with 1:200,000 epinephrine was injected submucosally above dentate line to promote hemostasis and facilitate mucosal dissection.

The endorectal mucosectomy was begun transanally 1cm cephalad to the dentate line (Fig. 1). On reaching the level of the peritoneal reflection, the muscle of the rectum was incised circumferentially to bring down the sigmoid colon through the anus (Fig. 2). The vessels that enter the colon were controlled with electrocautery only without the need to ligate it. We performed frozen section biopsies at the time of the procedure. It was taken from the dilated segment above the macroscopic transition zone.

Minilaparotomy through left lower quadrant hockey-stick incision was performed for 6 cases with long segment disease. The affected bowel was resected and the colo-anal anastomosis was done from below between the normally innervated bowel and the anal canal mucosa above the dentate line with 4/0 absorbable sutures.

The bladder catheter was removed on the 1st postoperative day. Oral feeding started with oral rehydration solution 24-48 hours postoperatively, followed by formula or breast feeding 24 hours later.

Outpatient visits was arranged weekly for one month and every 2 weeks for 3 months and then monthly thereafter. Rectal examination was performed two weeks after operation with the little finger for assessment of the anastomosis. Routine postoperative dilatation was not performed except in cases with anastomotic stricture.

RESULTS

Male to female ratio was 3:1. Two newborns had positive family history for Hirschsprung's disease.

The most common clinical manifestation of the studied group and age at operation are presented in tables (1,2) respectively. Associated anomalies included 4 patients with Down syndrome one of them had combined atrial and ventricular septal defects. Two other neonates had ventricular septal defect. One patient had hypospadias.

The barium enema with delayed x-ray films was diagnostic in 21 cases.

The transition zone was present at recto sigmoid junction in 13 cases, at rectum in 2 cases, at descending colon in 4 cases and at splenic flexure in 2 cases.

Frozen section biopsies confirmed the presence of ganglion cells in the proximal end of the resected colon in all cases: intraoperative blood loss was negligible and no blood transfusion was needed.

The mean operative time was 90±18 minutes including the time for frozen section biopsies. The length of hospital stay was 3 to 5 days. The time to resume oral feeding was ranging from 24-48 hours.

The length of the resected bowel varied between 14 and 35 cm.

The first per rectal examination was performed 2 weeks after operation with the little finger and only 2 patients required repeated dilatations due to anastomotic stricture.

There has been no recurrence of obstructive symptoms, one infant needed colostomy secondary to anastomotic leak manifested on the 4th postoperative day with fever, abdominal distension, ileus and confirmed by rectal examination that revealed disruption of the anterior half of the anastomosis.

One infant had prolapse of the pulled through colon on the 4th postoperative day which was reduced uneventfully.

Four patients required readmission for treatment of enterocolitis (admitted after operation, 2 weeks, 3 months, 10 months and 11 months). They were treated with saline rectal irrigation, no oral feeding and 3rd generation cephalosporins.

Perianal excoriation was the commonest postoperative complications occurring in 64.3% of cases and resolved within 8-14 weeks postoperatively.

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Table 1: The most common clinical manifestation of the studied group.

	No.	*%*
Delayed passage of meconium	26	92.9
Bilious vomiting	4	14.3
Abdominal distension	23	82
Enterocolitis	7	25

Table 2: The age at operation.

Age (days)	*No.*	*%*
5-7	2	7.1
8-14	5	17.9
15-21	9	32.1
22-30	12	42.9
Total	28	100

Table 3: Post operative complications of the studied group.

	No.	**%**
Anastomotic leak	1	3.6
Anastomotic stricture	2	7.1
Enterocolitis	4	14.3
Perianal excoriations	18	64.3
Prolapse of the pulled through colon	1	3.6

Table 4: Postoperative bowel movement frequency per day.

	No.
First month	5-8
Second month	4-6
Third month	4-6
Fourth month	3-5
Fifth month	3-4
Sixth month	2-4

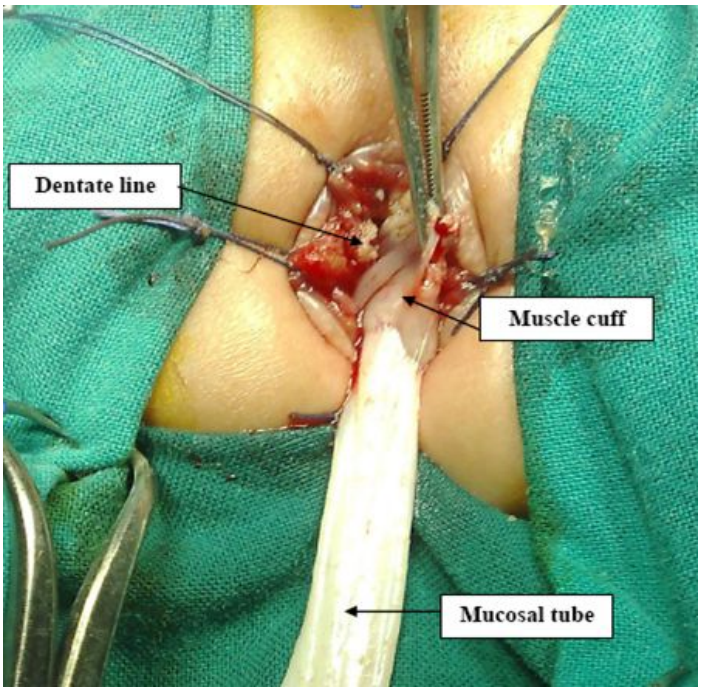

Fig. 1: transanal endo rectal mucosectomy for HD in a neonate

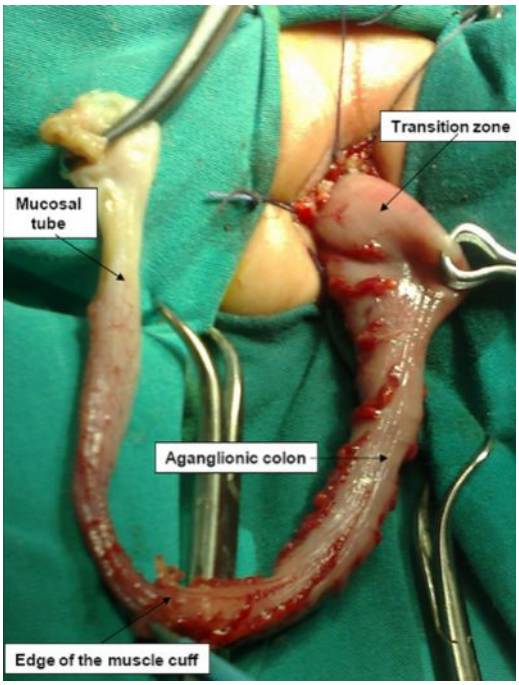

Fig. 2: Transanal pullthrough for the aganglionic colon

DISCUSSION

Ninety five percent of full term infants pass meconium in the first 24 hours of life but less than 10% of children with HD pass meconium during that time [5] in our series, 92.9% of cases experienced delayed passage of meconium.

Only 75% of neonates with HD will demonstrate a transition zone on barium enema. The absence of transitional zone doesn't rule out the diagnosis [18].

The initial descriptions of TEPT involved a long rectal cuff that reaches the peritoneal reflection but this cuff may constrict the pulled-through bowel, also the cuff may be rolled down into a ring during the pull-through so, some authors now prefer shorter distance for mucosectomy that come through the full-thickness of the rectum 2 to 3cm above the dentate line. This prevents, cuff stenosis and at the same time protect nerve, prostate and vagina from injury [5]. Moreover, many authors avoided mucosectomy and adopted transanal Swenson technique with successful preliminary results [19,20]. In our work, we continued to do mucosectomy till the peritoneal reflection without splitting the muscle cuff without recurrence of obstructive symptoms. This may be attributed to thinner muscle coat at this early age.

The mean operative time in our series was 90±18 minutes which is significantly shorter when compared with the Egyptian multi-center study of El-Halaby et al., [21], (120.2±27.8 minutes) and with that of Teeraratkul [15] (140 minutes). This difference may be caused by the younger age in our series.

Postoperative enterocolitis was noted in 14.2% of our series. This in contrast to the series conducted by Hadidi [22] where incidence of EC was 4.4% only, and the series of Gao et al [9] where the incidence was 6%. This higher incidence of enterocolitis in our series may be attributed to immaturity of the immune system in neonates [23]. In the absence of obstructive cause, enterocolitis is almost eliminated after the first 5 years of life [5]. The postoperative follow up in our series was short term and the last reported case with postoperative EC was presented 11 months after operation.

Apart from the number of motions per day (table 4) and assessment of continence state was not feasible as all the patients still use napkins. This issue is important due to lengthy anal stretching during mucosectomy. However, other studies showed no significant difference in manometric resting pressure between patients operated on transanally and those operated with abdominal endorectal pull-through [24].

We found that rectal mucosectomy in the neonatal period is easy and nearly bloodless with easier control of the blood supply of the pulled-through colon in reverse to mucosectomy in older children where the mucosal dissection is bloody due to thick muscle coat with inflamed, thickened mucosa and also ligation of the blood supply of the colon may be problematic due to thick mesentery of the long standing dilated colon [15].

Tannuri et al [25] proved that TEPT was simple and advantageous in newborns, in whom fixation of colon to retroperitoneum is looser which allows the resection of long segments of the descending colon through the anus, this in reverse to the more laborious procedure in older patients and in those with previous diversions or with deep rectal biopsies [26].

Like our research, there have been numerous articles reporting small series of patients undergoing the transanal pull-through [7-10,11,22,25]. On the other hand, two large series have been published with more than early follow up, the first of them was a multicenter study of 141 patients from North America and found that the transanal approach had a low complication rate, requires minimal analgesia and permits early feeding and discharge with 27% postoperative complication rate and 80.5% of the patients were felt to have normal bowel function by their care givers [27].

The second large series was also a multicenter study from Egypt and included 149 children who underwent transanal pullthrough with a similar results to the North American series [21]. About 50% of the North American study underwent surgery during the neonatal period but only 10% were neonatal in the Egyptian multi-center study.

In our series, laparotomy was needed in 6 cases due to long segment disease and no cases of the rest of the studied group (n=21) needed to be converted to laparotomy. This is in reverse to the Egyptian multicenter study [21] where the conversion rate was 2.7% due to tear of the mesenteric vessels and difficult mucosectomy. This difference may be attributed to easier mucosectomy and control of mesenteric vessels in the neonatal period. In this series, 64.3% of cases suffered from perianal excoriations which may be

attributed to more frequent motions and sensitive skin of the neonate. Similarly, this complication was present in more than half of the patients in the series of Wester and Rintala [17] especially for those whom were operated upon in the neonatal period.

It is well known that minimally invasive surgery reduces the incidence of intraabdominal adhesions [28], so non of our patients developed postoperative adhesions. Similarly most of the published series of TEPT did not show this complication especially when totally TEPT was adopted without laparotomy. Adhesive intestinal obstruction occurs after open pull-through for HD that ranges from 5% to 10% [14,29-31], especially when surgery is performed during neonatal period where the incidence of small bowel obstruction as a result of adhesions is higher [32].

A summary of published literature showing the results of transanal pull-through is seen in table (5)

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Table 5: Summary of the literature reporting the results of the transanal pull-through for Hirschsprung's disease.

Author	*No. of Patients*	*Time to Feeds (Hours)*	*% Requiring Narcotics*	*Hospital Stay (Hours)*	*Follow up (Months)*
De la Torre, 1998	5	24-36	-	-	11
Langer, 1999	9	24	55	48	6.5
Albanese, 1999	10	-	0	48	11
Langer, 2000	24	-	-	24	-
De la torre, 2000	10	24	-	-	21.3 ± 2
Gao, 2001	34	22.5	-	168 ± 48	10.5
Hollwarth, 2002	18	24	-	-	12.2
Teeratkul, 2003	8	24-48	-	144	3-12
Hadidi, 2003	68	48	-	72	21
Langer, 2003	141	36 ± 19.3	39	81.3 ± 30.8	20.2 ± 9.2
Ekema, 2003	15	24-48	epidural	144	7.1
Rintala, 2003	26	72	-	72	6
Wester, 2004	40	74.4	-	112	6
Elhalaby, 2004	149	-	-	115 ± 43.2	12
Tannuri et al,2008	35	36 ± 28	-	96.72 ± 88	-

CONCLUSION

The advantages of total transanal pull-through include bloodless easy mucosectomy, easier control of the blood supply of the colon, minimal resection of the dilated ganglionic part of the colon (dilatation and hypertrophy still mild), significant decrease in the need of analgesics in the immediate postoperative period, shorter hospital stay, decreased total cost, lower risk of adhesive intestinal obstruction and satisfactory cosmesis (no visible scars).

Rectosigmoid HD can be treated with the transanal endorectal pull-through technique but in case of long segment disease, laparotomy should be added to achieve an adequate colon dissection.

Long term outcome following transanal pull- through is still in its infancy especially for continence state that may be affected by the deleterious effect of the anal retraction on sphincter function.

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