Laparoscopic-aided transanal pull-through procedure for the management of Hirschsprung's disease: an observational prospective study for children older than 12 months Mohamed Rabea^{a,b}

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Received 24 March 2016 Accepted 15 April 2016

The Egyptian Journal of Surgery 2016, 35:348–356

Objective

The aim of the present study was to evaluate the surgical and functional outcome of the laparoscopic-aided transanal pull-through procedure for the management of Hirschsprung's disease (HD).

Patients and methods

The study included 17 patients older than 12 months. Diagnosis of HD relied on the inability of the patients to relax the anal internal sphincter in response to colonic extension on anorectal manometry. The laparoscopic part entailed transition zone identification, seromuscular biopsy for fresh frozen histopathology, and sigmoid and rectal mobilization up to 1 cm down the peritoneal reflection. The transanal part included mobilization of the rectal lower by 2 cm, resection till the ganglionic segment, and anastomosis. Patient outcome included evaluation of fecal consistency, frequency of soiling, and presence of perianal skin excoriation at 1, 3, and 6 months after the procedure. Colonic manometry and functional outcome evaluation according to qualitative clinical Holschneider scoring were carried out at 1 and 6 months after the procedure.

Results

One (5.9%) patient required open conversion for dissection of thick adhesions. Frequency of patients that passed formed stool and free of soiling increased progressively till the end of 6 months after the procedure. At 1 month after the procedure, six patients developed perianal skin excoriation, but all were free by the sixth postoperative (PO) month. Manometric pressure measures at 3 and 6 months after the procedure were significantly higher compared with the preoperative measures, with significant difference in favor of 6-month measures. At 6 months after the procedure, Holschneider scoring was significantly higher compared with that at 1 month, and 10 patients had a score of 14. No surgery-related complications were detected.

Conclusion

Laparoscopic-aided transanal pull-through is a feasible procedure for the management of children older than 12 months and who have HD. The procedure is safe with minimal PO complications, which are gradually resolved within 6 months PO.

Keywords:

anorectal manometry, functional outcome, Hirschsprung's disease, laparoscopic-aided transanal pull-through procedure

Egyptian J Surgery 35:348–356 © 2016 The Egyptian Journal of Surgery 1110-1121

Introduction

Hirschsprung's disease (HD) is a severe congenital anomaly of the enteric nervous system characterized by the absence of parasympathetic ganglion cells in the submucous and myenteric plexuses of the intestine, usually affecting the rectosigmoid portion of the large bowel. Distal innervation deficiency results from incomplete colonization of the bowel by enteric neural crest cells and results in functional intestinal obstruction [1].

HD remains the commonest cause of functional intestinal obstruction in children and contributes significantly to high morbidity and mortality. The

majority of patients present late, when the disease becomes complicated. Early diagnosis and timely definitive pull-through procedure are essential to decrease the morbidity and mortality associated with this disease [2].

Treatment of HD consists of surgical resection of the abnormal section of the colon, followed by reanastomosis. Definitive surgery for HD has been

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performed for many years using one of the techniques developed by Swenson, Duhamel, or Soave [3]. However, management of HD is still progressing due to application of minimally invasive procedures, minimizing the need for diversion stomas, reducing hospital stay, and postoperative (PO) morbidities and mortalities. Transanal endorectal pull-through (TERPT) for HD is a relatively safe and feasible procedure for neonates and infants. However, overstretching the anal sphincter and mesentery of the sigmoid colon might cause potential risk for impaired defecation function [4].

Introduction of laparoscopy as a part of the armamentarium for the management of HD permits obtaining biopsies so as to assure the level of aganglionosis, allows an adequate mobilization of the bowel with less trauma and bleeding, and, in addition, is cosmetically superior [5].

Various pull-through techniques, both open and laparoscopic, have been performed for HD and were compared for outcome; in Britain, the primary pullthrough, using an open Duhamel or laparoscopicassisted Soave–Boley technique, has become the operative strategy of choice for rectosigmoid HD, but marked variation in practice remains for the right-sided HD [6]. Open and laparoscopic Duhamel pull-through techniques were found to have similar outcomes with comparable operative times and hospital stay [7]. Laparoscopic-assisted endorectal pull-through procedure has become the standard treatment for HD in many centers around the world [8].

Recently, single-incision laparoscopic techniques have drawn more attention for their better cosmetic result. However, it is stressful for the surgeon in view of its low manipulability and poor visualization, causing clashing of instruments, especially in older children or patients with long-segment aganglionosis [4]. Clinical outcomes and ergonomic analysis of three laparoscopic approaches in the management of HD yielded the finding that laparoscopic approach should be selected according to the age, transition zone, and desired cosmetic result [9].

The current study aimed to evaluate the surgical and functional outcome of the laparoscopic-aided transanal pull-through procedure for the management of HD in children older than 12 months.

Patients and methods

The current prospective, observational study was conducted at El-Minia University Hospital (Egypt)

and Al-Eman Hospital (Riyadh, KSA) from June 2012 to March 2015 so as to allow a minimum follow-up period of 6 months for the last case operated upon. The study protocol was approved by the local ethical committee. An informed consent was obtained from the parents. All children with manifestations of chronic colonic obstruction were enrolled for clinical examination and investigations. Diagnosis of HD relied on the inability of the patients to relax their anal internal sphincter in response to colonic extension on anorectal manometry [10]. Inclusion criteria included age older than 12 months, chronic constipation secondary to HD, and no history surgical interference.

Collected clinical data included age, sex, weight, and frequency of presenting symptoms. Weight deficit, if any, was calculated versus the ideal weight-for-age using the US Center for Disease Control charts intended for babies and toddlers with an age range of 0–3 years [11]. All patients underwent abdominal radiograph in erect and supine positions and also barium studies to determine the site of probable aganglionic segment. Then, all patients underwent preoperative bowel preparation, including digital rectal stimulations and rectal irrigations using saline solution 20 ml/kg twice daily. All patients received prophylactic preoperative antibiotics as amoxicillin and clavulanic acid.

Operative procedure

All surgeries were conducted under general endotracheal anesthesia. Total body preparation was done for intraoperative change of position from laparoscopic to transanal part. Broad-spectrum antibiotic was given before skin incision. Operative procedure included two parts: a laparoscopic part and a transanal part. In the laparoscopic part, after placement of ports, the transition zone was identified (Fig. 1) and a seromuscular biopsy (Fig. 2) was obtained about 5 cm proximal to the transition zone frozen for fresh section histopathological examination so as to decide the level of pull-through. A window was made in the sigmoid mesentery and the sigmoid was mobilized. Proximal ganglionic bowel was mobilized, preserving the marginal arcade. The peritoneal reflection was sharply facilitate incised to dissection and mobilization of the aganglionic rectum. The rectum below the peritoneal reflection was minimally mobilized within 1 cm of peritoneal reflection (Fig. 3). After completion of laparoscopic dissection and assurance of hemostasis, the ports were left in situ and the patient's position was changed for transanal

Figure 1



The transition zone was identified.

Figure 2



Laparoscopic seromuscular biopsy.

Figure 3



Laparoscopic sigmoid and rectal mobilization.

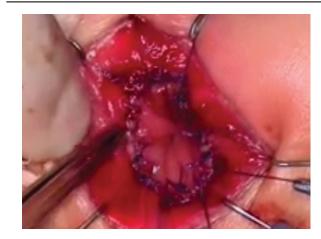
dissection of the remaining rectum. In the transanal part, as most of the rectal dissection was conducted laparoscopically, only the lower 2 cm of the rectum were mobilized transanally (Fig. 4). The anastomosis

Figure 4



Transanal dissection of lower rectal 2 cm.

Figure 5



Anastomosis was complete.

Figure 6



Anastomosis was complete.

was performed about 5 mm above the dentate line (Figs 5 and 6). The level of pull-through was at the biopsy-proven ganglionic segment. Once the anastomosis was completed, laparoscopy was performed again to check for the orientation of the pull-through bowel. Drain was placed and the port sites were closed.

Postoperative care

All patients received their immediate PO care in the neonatal intensive care unit (NICU). Patients were maintained on intravenous fluid therapy with continuous nasogastric decompression for at least 24 h. Broad-spectrum intravenous antibiotics were given for at least 48 h. Oral feeding was initiated when bowel sounds returned.

A patient was discharged when he or she could tolerate a full oral diet. Parents were taught to perform home dilatation with Hegar dilators. Rectal examination was performed 1 month PO. Patient's outcome was evaluated for fecal consistency, the frequency of soiling, and presence of perianal skin excoriation; the three items were evaluated at 1, 3, and 6 months after the procedure. Colonic manometry and functional outcome evaluation were carried out at 1 and 6 months after the procedure. Functional outcome was evaluated according to qualitative clinical Holschneider scoring [12], which included seven parameters scored on a three-point scale (Table 1), with a collective score of 14 points meaning normal bowel habits, 10-13 points meaning good (i.e. social continence with few limitation in social life), 5-9 points meaning fair (i.e. marked limitations in social life), and 0-4 points meaning poor bowel habits (i.e. total incontinence).

Statistical analysis

Obtained data were presented as mean±SD, numbers, and percentages. Results were analyzed using the oneway ANOVA with post-hoc Tukey's HSD test and the χ^2 -test. Statistical analysis was conducted using the SPSS (version 15 for Windows, 2006; SPSS Inc., Chicago, Illinois, USA) statistical package. A *P*-value less than 0.05 was considered statistically significant.

Results

The study included 17 patients fulfilling the inclusion criteria (12 boys and five girls), with a mean age of

19.8 \pm 6.5 years (range: 13–34 months). Mean body weight of the studied infants was 8.9 \pm 1.3 kg (range: 7–11 kg). Their weight deficit was 2.1 \pm 1.2 kg (range: 1–6 kg). Details of demographic data are shown in Table 2. All infants presented with chronic constipation, abdominal distension, and occasional passage of watery stool. However, all infants were accustomed to evacuating bowel once or twice weekly using enema. None of studied infants had previous operative interference.

Barium enema showed the typical picture of HD in 15 (88.2%) cases and demonstrated transition zone in the rectosigmoid region in 10 (58.8%) cases, in the sigmoid in three (17.6%) cases, and in the rectum in two (11.8%) cases. Two (11.8%) cases showed no transition zone on barium enema, but there was a marked rectocolic dilatation consistent with juxta-anal forms. Mean resting anorectal pressure and maximum internal sphincter pressure estimated on preoperative anorectal manometry were consistent

Data	Findings
Age (months)	
Strata	
12–18	8 (47.1)
19–24	4 (23.5)
25–30	4 (23.5)
>30	1 (5.9)
Total	19.8±6.5
Sex	
Male	12 (70.6)
Female	5 (29.4)
Weight (kg)	
Strata	
<10	12 (70.6)
≥10	5 (29.4)
Total	8.9±1.3
Deficit from ideal weight	
<2	6 (35.3)
2–4	10 (58.8)
>4	1 (5.9)
Total	2.1±1.2

Data are presented as n (%) and mean±SD.

	Parameters						
Scores	Frequency of defecation	Fecal consistency	Soiling	Sensitivity	Anorectal RP (mmHg)	Maximum squeezing MP (mmHg)	Adaptation reaction
2	Normal (1-2/day)	Normal	No	Normal	≥20–24	≥30	Normal
1	Often (3–5/day)	Soft	Stress/ diarrhea	Reduced	14–19	20–29	Small amplitude, shortened
0	Very often (>5/day)	Liquid	Constant	Missing	≤13	<20	Not detectable

MP, maximum pressure; RP, resting pressure.

with the diagnosis of HD and assured radiological findings.

All surgeries were conducted uneventfully without anesthesia or surgery-related mortalities. All patients underwent successful laparoscopic biopsy-taking, which were then examined by using the frozen section for assurance of the limit of the aganglionic part.

Laparoscopic part of the procedure was conducted free of complications, but one patient required open conversion for thick adhesions that hindered laparoscopic dissection, which consumed 25 min; the open procedure was completed uneventfully within a total operation theater time of 175 min and was considered a procedural failure with a rate of 5.9%. Mean total theater time for the remaining 16 patients was 159.3 ±15.5 min (range: 122–180 min). Intraoperative blood loss was minimal, with a mean amount of 30±11 ml (range: 20–60 ml). Mean length of resected colonic segment was 19.3±3.5 cm (range: 15–25 cm). Details of operative data are shown in Table 3.

All patients received their immediate PO care at the NICU. Mean time till resumption of oral intake was 32.9 ± 7.5 h (range: 20–48 h). Mean NICU stay was 46 ± 11 h (range: 30–70 h). All patients completed their NICU stay uneventfully. Mean total hospital stay was 4.9 ± 1.5 days (range: 3–6 days) for patients with successful laparoscopic part of the procedure, whereas the hospital stay for the patient who required conversion to open surgery was prolonged for 9 days. Details of operative data are shown in Table 4.

The frequency of patients who passed formed stool and free of soiling increased progressively till the end of 6 months after the procedure, with significant (P<0.05) difference compared with frequency determined at 1 and 3 months after the procedure, and significantly (P<0.05) higher frequency of formed stool passers at 6 months after the procedure

Table 3	Operative	data	of	studied	patients
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Data	Findings
Operative laparoscopy	
Successful	16 (94.1)
Failed	1 (5.9)
Operative time (min)	
Successful two part procedure	159.3±15.5
Open procedure	175
Total	160.3±15.5
Operative blood loss (ml)	30±11
Length of resected colonic segment (cm)	19.3±3.5

Data are presented as n (%) and mean±SD.

compared with at 3 months after the procedure. Six patients developed perianal skin excoriation, detected at 1 month after the procedure; three were still excoriated at 3 months after the procedure, but none still had excoriation at the sixth PO month (Table 5).

Mean resting anorectal pressure and maximum internal sphincter pressure at maximum squeeze estimated at 3 and 6 months PO were significantly (P<0.05) higher compared with the preoperative pressure estimates, with significantly (P<0.05) higher pressure measures at 6 months compared with the measures estimated at 3 months PO (Fig. 7).

Qualitative clinical Holschneider scoring of studied patients at 6 months PO was significantly higher compared with the 1-month scoring (Table 6 and Fig. 8). Patients' distribution among the strata of Holschneider scoring was significantly higher at 6 months compared with the 1-month distribution and 10 patients at 6 months, whereas only one patient at 1 month had a score of 14 (Fig. 9). No surgery-related complications or wound dehiscence were detected.

Discussion

The current selective study included children with HD older than 12 months to allow anorectal manometric diagnosis and follow-up depending on the fact that anorectal manometry was carried out only in patients at least 12 months old because the relax reflex of the anal internal sphincter may not be developed in younger infants [13]. Selection of such an age-group aimed also

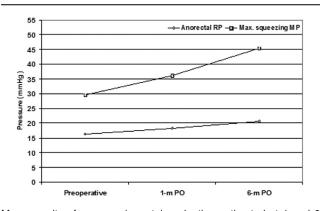
Table 4 Immediate	postoperative	data of stu	died patients
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Data	Findings
Time till resumption of oral intake (h)	
<24	2 (11.9)
24–35	8 (47.1)
36–48	7 (41)
Total	32.9±7.5
NICU stay (h)	
<36	2 (11.9)
36–48	12 (70.3)
>48–60	1 (5.9)
>60	2 (11.9)
Total	46±11
Hospital stay (days)	
3–4	5 (29.4)
5–6	11 (64.7)
>6	1 (5.9)
Total	5.2±1.3

Data are presented as n (%) and mean±SD. NICU, neonatal intensive care unit.

		Stool c	Stool consistency			Occurrence of soiling	br	Presence	Presence of excoriated perianal skin	rianal skin
	Liquid	Loose	Formed	Р	Yes	No	Р	Yes	No	Ρ
1 month	4 (23.5)	5 (29.4)	8 (47.1)	I	5 (29.4)	12 (70.6)	I	6 (35.3)	11 (64.7)	I
3 months	1 (5.9)	3 (17.6)	14 (82.4)	$P_{1}=0.001$	3 (17.6)	14 (82.4)	$P_{1} > 0.05$	3 (17.6)	14 (82.4)	$P_{1}=0.042$
6 months	0	1 (5.9)	16 (94.1)	P ₁ =0.001	1 (5.9)	17 (94.1)	$P_{1} > 0.05$	0	17 (100)	$P_{1}=0.001$
				$P_2 = 0.045$			$P_2 = 0.011$			$P_2 = 0.034$





Mean results of monomeric rectal evaluation estimated at 1 and 6 months postoperatively (PO) compared with preoperative results. MP, maximum pressure.

to provide studied patients with the best chance for better surgical and functional outcomes. In support of age-dependence of outcome, Sun *et al.* [14] found PO perianal erosion, enterocolitis, and soiling to be significantly higher in HD infants undergoing operation within 3 months compared with those more than 3 months of age.

The applied operative procedure consisted of two parts (a laparoscopic part and a transanal part). The laparoscopic part, first, entailed laparoscopic exploration for the aganglionic segment and seromuscular biopsy-taking for the definition of the proximal level of the aganglionic segment, depending on the histological examination of the fresh frozen section. In line with the reliance on the histological examination of the fresh frozen section, Takawira et al. [15] compared contrast enema, anorectal manometry, and biopsy with histology for the diagnosis of HD and found that hematoxylin and eosin-stained fresh frozen sections remain the criterion standard for its highest sensitivity and specificity.

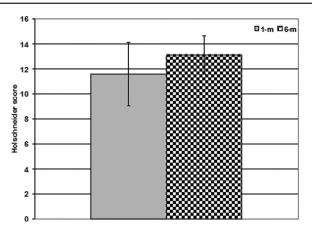
Intraoperative fresh frozen section histological examination allowed proper identification of the normal ganglionic level so that no remnant aganglionosis was missed; in support of such a policy, throughout the follow-up period, no patient developed recurrent constipation. Moreover, Takahashi *et al.* [16] and Bonnard *et al.* [17] found that laparoscopic biopsy-taking can aid diagnosis and decision-making in HD cases.

Second, laparoscopy allowed dissection of the colon and rectum up to 1 cm below the peritoneal reflection, so that the transanal part was accomplished easily and uneventfully owing to the minimal length required to

reaction	
1.8±0.6	11.6±2.5
1.9±0.3	13.1±1.5*
	1.8±0.6

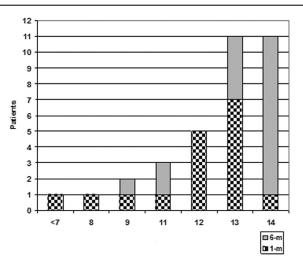
MP, maximum pressure; RP, resting pressure. *Significant difference.

Figure 8



Mean±SD of qualitative clinical Holschneider scoring of studied patients determined at 1 month and 6 months.

Figure 9



Patients' distribution among strata of Holschneider scoring determined at 1 and 6 months postoperatively.

be dissected. Laparoscopy also assured hemostasis and, finally, helped in checking for orientation of the pull-through bowel.

Thus, the major part of the surgical burden was applied laparoscopically and was completed uneventfully in all cases apart from one case in which dense adhesions hindered dissection and was converted to laparotomy with a conversion rate of 5.9%. These data indicated the feasibility and safety of the laparoscopic part of the procedure. In addition, meticulous under-vision dissection and minimal tissue trauma reduced the PO catabolic stage, thus allowing short duration of NICU stay and time till resumption of oral intake.

The reported advantages of laparoscopy for the management of HD cases were in agreement with those reported by Ksia *et al.* [18], who retrospectively studied 20 patients older than 2 years who underwent a transanal Soave one-stage endorectal pull-through procedure for HD, and concluded that laparoscopy may be necessary whenever there are difficulties in the pull-through.

In their study, van de Ven et al. [19] reported resection of significantly longer colon by using the TERPT within significantly shorter operative time compared with laparoscopically-assisted transanal endorectal pull-through (L-TERPT), but found a significant association between length of resection and PO obstructive symptoms, and also reported nonsignificantly higher frequency of colonic torsion after TERPT compared with L-TERPT. In addition, Gosemann et al. [20] documented that L-TERPT represents a valid option in the treatment of HD and might have some advantages over the open techniques. Moreover, Mathur et al. [21] found that the extent of laparoscopic mobilization of rectum does not appear to be a factor deciding the outcomes, but laparoscopic assistance can be used to maximize the benefits of Swenson type of operation and a transanal pull-through.

The advent of laparoscopic application for the management of HD could be another support for the advantages reported by the current study, whereas Tang *et al.* [22] retrospectively documented that in selected HD patients, the single-incision laparoscopic endorectal pull-through (SILEP) was safe and technically feasible in experienced hands, with similar operative results compared with conventional laparoscopic endorectal pull-through, but SILEP is more difficult if the transition zone is higher than the rectosigmoid. Moreover, Aubdoollah *et al.* [4] reported no major intraoperative complication for hybrid SILEP; PO,

perianal excoriation was the main early complication, which occurred in nine patients, and enterocolitis in two patients, but no anastomotic leak occurred.

As another form of support for advantages of laparoscopy, Tang *et al.* [23] compared laparoscopic surgery with traditional perioperative management versus fast-track management for infants with HD and reported no significant differences in intraoperative blood loss and operative time, and despite the earlier recovery of bowel movement with fast-track management, the difference was not statistically significant, with similar PO complications and recovery conditions during 4 weeks of follow-up.

In addition, Thomson et al. [24] conducted a metaanalysis for the outcomes following totally TERPT versus TERPT with any form of laparoscopic assistance for infants with uncomplicated HD, and found no significant differences concerning PO enterocolitis, fecal incontinence, or constipation. Recently, Scholfield and Ram [25] conducted a meta-analysis including 11 articles comparing open and versus laparoscopic Duhamel procedures for HD and reported significantly greater incidence of soiling/ incontinence, further surgery, longer hospital stay, and time to oral feed, but significantly shorter operative time with open procedure compared with the laparoscopic procedure, whereas the incidence of enterocolitis and constipation was nonsignificant.

Conclusion

The obtained results and review of the literature allowed concluding that laparoscopic-assisted transanal pullthrough is a feasible procedure for the management of children older than 12 months and with uncomplicated HD. The procedure is safe with minimal PO complications that are gradually but progressively resolved within 6 PO months. However, wider-scale randomized comparative studies are mandatory for establishing such outcome.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

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