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ORIGINAL ARTICLE

Bowel function and fecal continence after Soave's trans-anal endorectal pull-through for Hirschsprung's disease: a local experience

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Abstract The aim of this study was to evaluate the postoperative clinical outcome, colorectal function, and fecal continence score after Soave's transanal endorectal pull-through surgery (TERPT) for Hirschsprung's disease (HD) comparing them in preschool and school children with the results of younger children. This comparative retrospective study was done on 40 HD children treated over a period of 8 years from January 2001 to December 2008. Patients were classified into two equal groups according to their age: group I ($n = 20$) included children with age <6 months up to 42 months, and group II ($n = 20$) included children from 3.5 years up to 13 years. Demographic, clinical data, preoperative investigations, operative records, postoperative outcome and follow-up including defecation problems, fecal continence score rate (FCSR), anal manometry and electromyography were all reviewed. Obtained data were statistically analyzed using SPSS. Forty patients were included in this study, 28 males and 12 females with the male to female ratio of 2.3:1. The median age of the studied patients in group I was 8.9 months, while in group II, the median age was 65.95 months. The postoperative follow-up period ranged from 18 to 24 months in group I with a mean of 21 months, while it ranged from 2 to 26 months in group II. In group I, most of children showed no abnormal defecation problems, 16 patients had excellent FCSR, 4 were having good FCSR and no poor continence score rate, while 3 patients suffered from constipation. Meanwhile, in group II, 15 patients showed excellent FCSR in 10 patients and 5 with good FCSR. While the rest of patients suffered from different abnormal defecation behavior that was constipation in 5 patients. The remaining 5 patients suffered from continence problems varying from fair in 3 patients (20%), with the remaining 2 patients having a poor continence score rate. It can be concluded that TERPT can be performed with some difficulties in older children; yet, the follow-up results are statistically low when compared with those patients who had undergone the operation at younger age.

Keywords Hirschsprung's disease · Bowel function · Fecal continence · Soave's TERPT

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Introduction

Transanal endorectal pull-through operation (TERPT) for Hirschsprung's disease (HD) has been widely performed because it has a low degree of invasiveness [1]. Recently, several reports were published to compare long-term bowel function for TERPT procedure alone or to conventional abdominal procedure [2, 3]. Although, most patients with operated HD have good functional results in adult hood [4, 5], a majority have problems with constipation and incontinence in childhood [6]. In previously published papers, the bowel function evaluations were based on the clinical evaluation [2, 3]. The aim of our current work is to follow up those HD patients who underwent TERPT procedure at different ages, as regards to the clinical outcome, bowel function, and fecal continence. This follow-up was

not only based on clinical evaluation but other investigatory tools were included such as electromyography (EMG), postoperative anal manometry (AM), and magnetic resonance imaging (MRI) in order to investigate the effect of age at the time of surgery on the late bowel function on those patients.

This work aimed at evaluating the postoperative clinical outcome, colorectal function, and fecal continence score after TERPT.

Patients and methods

This comparative retrospective study was done on 40 HD children who were treated at the Division of Pediatric Surgery, Ismailia, Egypt, over a period of 8 years from January 2001 to December 2008. These patients were randomly selected using systematic random sample. All our patients were having a classical form of HD disease with the narrow segment level at the recto-sigmoid junction or below. Cases of long segment and also ultra-short HD were excluded from this study.

They were equally divided into two groups (20 each) based on the age at the time of surgery. In group I ($n = 20$), patients' age ranged from 6 months to 3.5 years, however, in group II ($n = 20$), the patients' age was between 3.5 years and 13 years. After approval of the hospital ethical committee, we have reviewed all the charts of the patients. They were thoroughly studied regarding demographic data including age of presentation, sex, history of consanguinity, similar cases in the family, clinical data including natal and postnatal history, delayed passage of meconium, neonatal intestinal obstruction and/or chronic constipation and fecal soiling and/or encopresis in older children.

Preoperative investigations were studied including conventional radiological investigations or rectal biopsies. The numbers of ostomies pre-definitive surgical procedures, operative data including the used technique, mean operative time and intraoperative difficulties or complications if any were recorded. The length of excised specimen, requirement of blood transfusion, onset of oral intake, length of stay, postoperative complications such as perineal excoriation, stricture, enterocolitis (EC), perineal or pelvic infection, functional outcome and need of a secondary surgery were investigated. We have defined the EC as a clinical condition with abdominal distention, general sickness, diarrhea, pyrexia and pain.

Postoperative outcome and follow-up were also studied including some common complications such as EC, and fecal incontinence. We called all the patients over phone asking for bowel habits, any experience of perineal excoriation, stricture, EC symptoms after surgery and any other health problem subsequent to the discharge from the hospital. They were reviewed at 1 week and 1 month after discharge. The parents or the caretakers were informed about the symptoms of the EC, and they have been told to admit the patient to the next health center at the earliest convenience in case of any bowel problem. Fecal continence score rate (FCSR) using the Wingspread scoring system [7–9] was applied. This scoring system has been widely used for postoperative continence evaluation in patients with anorectal anomalies; yet, it can easily and effectively evaluate continence post HD surgery.

In this score, an excellent or very good score means a totally continent or very occasional stress-related soiling of underclothes without constipation and toilet trained with no medication. Good score was considered if patient rarely soils except during exercise or constipation that is amenable to management with medication. While fair score means intermittent soiling, urge incontinence, frequent loose stools or constipation that require enema. Poor score means constant fecal soiling and smearing and constipation only responsive to enema. A thorough clinical examination was applied to all patients. AM, EMG and magnetic resonance imaging (MRI) were applied for those who showed an FCSR below good.

Postoperative AM was performed to evaluate the maximum resting pressure where a normal value was considered to range between 50 and 80 mmHg, a normal maximum squeeze pressure between 90 and 180 mmHg. Low maximum resting and squeeze pressure indicate weak anal sphincter muscles. All of EMG results were also thoroughly reviewed. MRI was performed to those patients with poor and fair Wingspread score, to investigate the muscular cause of the incontinence among them. Collected data were recorded in a semi-structured form. The Statistical Package for Social Sciences (SPSS) version 17 was used for both data tabulation and analysis. It was presented as appropriate in the form of frequencies and percentages mean and SD, Chi-Square test was used for qualitative data, and Student t test was used for quantitative data. The level of significance selected for this study is $P \leq 0.05$.

Results

Forty patients were included in this study; 28 males and 12 females with the male to female ratio of 2.3:1.

In group I ($n = 20$), patients' age was between 6 and 35.7 months with a median age of 8.9 months. However, in group II, the age varied from 36 to 159 months with a median of 65.95 months (Table 1).

Preoperative clinical data of our studied groups were summarized in Table 2.

Table 1 Demographic data of studied HD patients

HD	Group I ($n = 20$)	Group II ($n = 20$)
Male	15 (75%)	13 (65%)
Female	5 (25%)	7 (35%)
Mean age (mean ± SD):	14.02 ± 10.3	69.9 ± 32
Minimum	5	36
Maximum	35.7	156
Median age	8.9	65.95
+ve family history of HD	3 (15%)	2 (10%)
+ve consanguinity	9 (45%)	7 (35%)

The postoperative follow-up period ranged from 18 to 24 months in group I with a mean of 21 months, while it ranged from 2 to 26 months in group II.

Anorectal manometric data is shown in (Table 3). The maximum resting pressure is normally ranging from 50 to 80 mm Hg; the normal maximum squeeze pressure ranges from 90 to 180 mm Hg. The low maximum resting and squeeze pressure indicate weak anal sphincter muscles. We recorded a median maximum resting pressure of 62 and 51.4 for groups I and II, respectively. The median maximum squeeze pressure was 139 and 80.5 for groups I and II, respectively.

Our EMG map of the sphincter did not detect any silent areas, denoting no postoperative sphincter damage.

In group I, most of children showed no abnormal defecation problems: 16 patients had excellent FCSR, 4 were having good FCSR and no poor continence score rate, while 3 patients suffered of constipation.

Meanwhile, in group II, 15 patients showed excellent FCSR in 10 patients and 5 with good FCSR, while the rest

Table 2 Showing analysis of the different studied items in both groups with their statistical significance

Item	Group I	Group II	Statistical significance
Patient number	20	20	–
Patient's age (months) (mean ± SD)	14.02 ± 10.3	69.9 ± 32	$P < 0.001$
Presenting symptoms:			
1. Delayed passage of meconium	15 (70%)	–	–
2. Recurrent constipation	5 (30%)	20 (100%)	$P < 0.001$
3. Preopreative enterocolitis	2 (10%)	7 (35%)	$P = 0.127$
4. Abdominal distension	16 (80%)	20 (100%)	$P = 0.053$
Transition zone (TZ)	14 (70%)	17 (85%)	$P = 0.225$
Mean operative time:			
(Mean ± SD)	106 ± 16.8	163.7 ± 24.2	$P < 0.01$
Minimum	80	120	
Maximum	130	200	
Median	107	168	
Postoperative hospital stay:			
(Mean ± SD)	4.4 ± 1.6	11.3 ± 3.1	$P < 0.001$
Minimum	2	7	
Maximum	7	16	
Median	4.3	11.2	
Postoperative complication:			
1. Need blood transfusion	0 (0.0%)	2 (10%)	$P = 0.244$
2. Postoperative fever	2 (10%)	6 (30%)	$P = 0.118$
3. Prolonged postoperative ileus	No (0%)	3 (15%)	$P = 0.115$
4. Duration of postoperative soiling and fecal incontinence			
(Mean ± SD)	36.98 ± 8.8	87.38 ± 7.4	$P < 0.001$
Minimum	25	75	
Maximum	52	100	
Median	35.8	87	
5. Perianal excoriation	1 (5%)	7 (35%)	$P = 0.022$
6. Postoperative enterocolitis	No (0%)	3 (15%)	$P = 0.115$
7. Postoperative stricture	No (0%)	3 (15%)	$P = 0.115$

$P < 0.05$ is significant

Table 3 Results of postoperative AM ($n = 40$)

	Group I (%)	Group II (%)	*P* value
Maximum resting pressure			
Normal	19 (45%)	16 (80%)	0.342
Abnormal	1 (5%)	4 (20%)	
Mean ± SD	63.3 ± 6.4	49 ± 5.3	
Minimum	50	47	
Maximum	75	63	
Median	62	51.4	
Maximum squeeze pressure			
Normal	19 (45%)	16 (80%)	0.342
Abnormal	1 (5%)	4 (20%)	
Mean ± SD	135.95 ± 21.8	83.4 ± 9.6	
Minimum	92	70	
Maximum	167	98.9	
Median	139	80.5	

No statistically significant difference ($P > 0.05$)

of patients suffered of different abnormal defecation behavior that were constipation in 5 patients. The remaining 5 patients suffered of continence problems varying from fair in 3 patients (20%), with the remaining 2 patients having a poor continence score rate. There was a statically significant difference between the 2 groups regarding a better FCSR in group I compared to group II; *P* value = 0.001 (Table 4). On the other hand, none of the patients, even those with poor or fair continence score rate, showed any abnormal MRI findings of the pelvic floor or anorectal muscle complex.

Discussion

The one-stage transanal endorectal pull-through TERPT has become a very popular surgical option in treatment of HD during the last 17 years since it was firstly described [1, 9–13].

This procedure is less invasive, resulting in better esthetic and cause less postoperative pain and complications than laparotomy. Short segment aganglionosis occurring distal to the sigmoid colon is a good indication for TERPT without laparoscopic assistance or laparotomy [10].

Postoperative outcomes of this procedure have not yet been adequately assessed regarding the postoperative AM and/or EMG. It has not been reported that the incidence of constipation of fecal incontinence is not always satisfactory after this operation [6, 13–15].

In our current study, we investigated the postoperative continence score rate after TERPT in 2 different age groups, and we found a statistically significant difference regarding this issue with better results if the operation was performed at a lower age. Moreover, it was technically easier and more feasible if TERPT was done in younger age group.

From literature review, many case series with TERPT had excellent short—and medium—term clinical and functional results. Furthermore, there are some fatal cases after TERPT in some series [16–18], yet, we reported no mortality in our studied cases. Our follow-up period ranged from 18 to 24 months in group I, while it ranged from 2 to 26 months in group II. This could be compared to others published data where they recorded a range of follow-up period from 6 months to 5 years with a mean of 28 months on series that were done on 21 patients who underwent TERPT at a young age group ranging from 26 days to 6 years [19]. Their data also agreed on what we have done preoperatively to our patients regarding the use of contrast study of the colon before surgery, the use of preoperative bowel preparation. However, in our series blood transfusion was needed in two elder patients due to intraoperative bleeding compared to the need of blood transfusion in other series [11, 19]. Our mean operative time was significantly shorter when the operation was performed in a younger age with less operative complications, postoperative hospital stay and early oral feeding. This may be attributed to the

Table 4 Relation between Wingspread FCSR system for incontinence and age ($n = 40$)

Wingspread scoring system	Group I		Group II					Total	*P* value
	Surg.age <0.5 months (%)	1 year (%)	3.5 years (%)	4 years (%)	4.5 years (%)	5 years (%)	6 years (%)		
Excellent	13 (26)	3 (6)	7 (14)	2 (4)	1 (2)	0 (0)	0 (0)	26	**0.001**
Good	2 (4)	2 (4)	3 (6)	1 (2)	1 (2)	0 (0)	0 (0)	9	
Fair	0 (0)	0 (0)	0 (0)	0 (0)	1 (2)	1 (2)	1 (2)	3	
Poor	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	1 (2)	1 (2)	2	
Total	15	5	10	3	3	2	2	40	

Surg. age age at surgery

Bold value is statistically significant

fact that dissection is easier and intraoperative bleeding is less in younger patients. Another factor is that older children who suffered a long time chronic constipation might have a hugely dilated colon that is difficult to dissect as well as the recurrent enterocolitis in this age; it leads to adhesions and difficult surgery.

Some authors found no statistical difference in the mean operation time between different groups who underwent abdominal pull-through TERPT and laparoscopically assisted TERPT [20].

This was supported by previous published data though it was contradicted in our series when the operation was done in elder age group where 2 patients showed prolonged ileus that did not permit us to start early oral feeding. Perianal excoriation was seen in only one patient of our younger children and 7 patients in the elder group with a total percentage of 20%. Such data could be compared to others who reported 11 patients out of total 21 (52.3%) who suffered of postoperative mild perianal excoriation that responded to local ointments. Some other published data of a multi centric study done in the same environment showed 48 patients out of total 149 (32.2%) with perianal excoriation [11]. Another study reported perianal excoriation to occur in 15 out of a total 141 patients (10.6%) [17]. Postoperative ileus was seen in three patients of group II, opposed by two published data on similar age group [21–23]. Yet, our negative recorded data of intestinal obstruction in group I coincided with many published data that showed no obstruction in younger children [10, 11, 21]. On the other hand, in our study, the percentage of constipation in groups I and II were 15 and 30%, respectively. This data was opposed by other data that showed no constipation in a series of 21 younger children, though, they reported four children to have a benign postoperative diarrhea with dressed having normal bowel habits. Nevertheless, some others did agree with our data of constipation [11, 17, 23–26]. Our data showed that relatively elder children are more amenable to complications whether preoperative, difficult operation and significantly more pronounced postoperative complications coinciding with different data published in the literature stressing on bowel function and fecal incontinence [27].

In our current study, EMG and AM though not feasible in younger children were tried in those around the age of 3 years or even less and they proved to be effective in our series.

AM was used for all patients in order to follow-up their defecation with a mean resting pressure of 63.3 ± 6.4 and 49 ± 5.3 in groups I and II, respectively. Similarly, this was published in the literature [28] where the average of anal resting pressure ranged from 15 to 52.3 of post TERPT patients who had a maximum age of 4.4 years. Reported incontinence was not associated with anatomical disruption as noted by results of MRI but it was associated with abnormal EMG results denoting that the reporting fecal incontinence post HD surgery may not be due to surgical reasons.

EMG was done for our patients who did show a fair or unsatisfactory FCSR to elucidate if TERPT does affect the muscles. The results showed abnormal EMG results denoting that the reporting fecal incontinence post HD surgery is not due to surgical reasons. In addition, we can state that reported incontinence is not associated with anatomical disruption as noted by results of MRI.

It can be concluded that TERPT operation is a very good option in treating most classic cases of HD with excellent results and amenability in younger children with difficulty in older children although it may be feasible. Elder children follow-up fecal continence results are statistically low compared with those patients who had undergone the operation at younger age.

Therefore, it may be better to start diagnosis and surgery for HD patients at younger age especially when TERPT would be the surgical option as the incidence of preoperative recurrent enterocolitis and chronic constipations will be at its minimum leading to an easy surgery with lesser complications. Health education and counseling with parents are essential to clarify the importance of performing the surgery as early as possible to avoid postoperative complications especially fecal incontinence. This may be considered as a part of a health programs in developing countries like ours.

Conflict of interest None.

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