Transanal Swenson Pullthrough in Hirschsprung Disease: Ain Shams University Experience

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ABSTRACT

Introduction: *Hirschsprung's disease (HD) constitutes a serious developmental disorder of alimentary system resulting in functional obstruction secondary to absence of ganglion cells in distal region of intestinal wall. Inappropriate treatment of the disease entails a risk of life-threatening sequalae including enterocolitis in neonates or failure to thrive with defecation problems in older children. Several creative approaches have been described in the surgical management of Hirschsprung's disease.* ***Aim of this Study:*** *is to report our experience and initial results of the Transanal Swenson PullThrough (TSPT) technique performed on 35 Hirschsprung patients because there are technical aspects in this operation that differ from the method originally described by Swenson.* ***Materials and Methods:*** *Our study includes 35 patients with HD (22 boys and 13 girls) who underwent TSPT at Ain Shams University Hospitals with an age range between 3 months and 3 years within a time range of 5 years from January 2011 till December 2015.* ***Results:*** *Postoperative complications included anastomotic stricture in 2 patients (5.7 %), enterocolitis in 3 patients (8.5 %), urethral injury in 2 patients (5.7 %),Perianal excoriation in 3 patients (8.5 %) and fecal incontinence in 1 patient (2.8 %). The overall complication rate is 11/35 (31.4 %). There were no anastomotic leaks or bowel obstruction. Defecation rate of 1–3 times per day was noted in 80% (28/35) of cases within six months of surgery and 14.2% (5/35) at 1 to 2 years post surgery. There were no mortalities in the series.* ***Discussion:*** *Dissection of the aganglionic segment was pretty stressful as we were aiming at confining the dissection plane as close to the bowel wall as possible, to avoid injuring thepresacral plexus and anterior structure of the pelvis. Another critical issue was related to the evaluation of the transition zone. We had comparable results and post operative complications with other studies, yet less operative time.* ***Conclusion:*** *Our initial results are really promising. By applying a meticulous dissection strategy, injury to the surrounding structures can be avoided and blood loss can be minimized. So we can conclude that this technique has acceptable short-term outcomes. A long-term follow-up would still be necessary to assess the real impact of the technique on children's wellbeing as they grow up.*

Key Words: *Transanal Swenson pull-through, Hirschsprung disease, one-stage transanal procedure, transanal rectosigmoidectomy, trophic feeding, post-operative encopresis, enterocolitis.*

INTRODUCTION

Hirschsprung's disease (HD) constitutes a serious developmental disorder of alimentary system resulting in functional obstruction secondary to absence of ganglion cells in distal region of intestinal wall.[1]

It affects one in every 5,000 newborns. The diagnosis is often suspected when a newborn fails to pass meconium in the first 48 hours of life, has abdominal distension, or has vomiting. In older children, a history of chronic constipation may be the presenting complaint. The diagnosis is suspected by a barium enema suggesting a transition point and confirmed by rectal biopsy showing the absence of ganglion cells.[2]

Inappropriate treatment of the disease entails a risk of life-threatening sequalae including enterocolitis in neonates or failure to thrive with defecation problems in older children.[3]

Since its first recognition in 1888, many techniques of surgery for the treatment of Hirschsprung's disease (HD) have been proposed. The surgical management of the recto-sigmoid HD is rapidly changing from the three-staged procedure to a single-stage, transanal, pull-through procedure. This evolution aims at leaving no scars and reducing the cost, pain, hospital stay, and the morbidity associated with the staged procedures. [4]

Transanal Swenson pull-through operation (TSPT) is the earliest modification of the transanal approach that completely removes the aganglionic bowel by dissecting full thickness of the rectum.A number of definitive procedures have shown excellent results when performed by experienced surgeons. Currently, the most

common repairs performed are the Swenson, Duhamel, and Soave procedures. [5]

In any elective operation for Hirschsprung disease, a robust preoperative colon cleanse must be performed.[6] Intraoperatively, histological examination of a frozen-section biopsy must confirm the presence of ganglion cells at the proximal margin of bowel intended for anastomosis. A meta-analysis performed by Friedmacher and Puri in 2011 reported that residual aganglionosis and transition-zone tissue account for persistent bowel symptoms in one third of patients undergoing a second, corrective pull-through procedure.[7]

The Swenson procedure was the original pull-through procedure used to treat Hirschsprung disease. The aganglionic segment is resected down to the sigmoid colon and rectum, and an oblique anastomosis is performed between the normal colon and the low rectum.[8] The Duhamel procedure was first described in 1956 as a modification to the Swenson procedure. A retrorectal approach is used, and a significant segment of aganglionic rectum is retained. The aganglionic bowel is resected down to the rectum, and the rectum is oversewn. The proximal bowel is then brought through the retrorectal space (between rectum and sacrum), and an end-to-side anastomosis is performed with the remaining rectum.[9]

As for the Soave procedure, it was introduced in the 1960s. The mucosa and submucosa of the rectum are resected, and the original operation did not include a formal anastomosis, relying on scar tissue formation between the pull-through segment and the surrounding aganglionic bowel. The procedure has been modified by Boley to include an endorectal mucosectomy, which leaves a long muscular cuff, that is usually split posteriorly with an end-to-end coloanal anastomosis.[4] The problem is that this long muscular cuff that is left behind causes obstruction because according to the concept of this procedure, some aganglionic tissue is still retained in the rectal cuff. [10] In addition, a technique-related complication, such as procedural difficulty in pulling down the ganglionic bowel due to limited volume of the remaining seromuscular cuff have been recently reported. [11]

For patients with extremely short-segment Hirschsprung disease, anorectal myomectomy is an alternative surgical option where a 1-cm-wide strip of extramucosal rectal wall is excised, beginning immediately proximal to the dentate line and extending to the normal ganglionic rectum. The mucosa and submucosa are preserved and closed.[12]

Patients with total colonic involvement require modified procedures to exclude the aganglionic colon while preserving maximal absorptive epithelium. The goal of these procedures is to bypass dysfunctional bowel while maximizing the chance of postoperative nutritional function and growth.[12]

Transanal Swenson pull-through operation (TSPT)have been described in which no intra-abdominal dissection is performed**.** [13] The entire procedure is performed transanally in a manner similar to perineal rectosigmoidectomy.The mucosa, submucosa and muscularisis incised circumferentially above the dentate line, and the dissection is carried external to the rectal wall proximally until the transition zone is identified. Upon confirmation of ganglion cells on frozen section, the aganglionic bowel is resected and an anastomosis is performed.[14]

Several other creative approaches have been described, including a modification of the transanal approach with transabdominal open or laparoscopic assistance, single-incision laparoscopic endorectal pull-through (SILEP), and natural orifice transluminal endoscopic surgery (NOTES).[15]

A laparoscopic approach to the surgical treatment of Hirschsprung disease was first described in 1999 by Georgeson.[16]The transition zone is first identified laparoscopically, after which the rectum is mobilized below the peritoneal reflection. A transanal full thickness dissection is performed, then the rectum and aganglionic bowel is prolapsed through the anus.After excision of the aganglionic segment, the healthy proximal bowel is anastomosed to the anus 1cm above the dentate line. Functional outcomes of this laparoscopic approach appear to be equivalent to open techniques based on short-term results.[17,18]

Outcomes of the TSPT procedure have been similar to open single-stage approaches, and analgesia requirements and hospital stays are decreased.[19] Recent studies also report lower rates of postoperative incontinence and shorter

operating times in transanal pull-through procedures.[19]

Aim of the work

The aim of this study is to report our experience and initial results of the Transanal Swenson PullThrough (TSPT) technique performed on 35 Hirschsprung patients because there are technical aspects in this operation that differ from the method originally described by Swenson.

MATERIALS AND METHODS

Our study includes 35 patients with HD (22 boys and 13 girls) who underwent TSPT at Ain Shams University Hospitals with an age range between 3 months and 3 years within a time range of 5 years from January 2011 till December 2015. A written consent was acquired from the patients' guardians after having explained the procedure to them. Patients with total colonic aganglionosis,life-threatening anomalies, deteriorating general health, and severe enterocolitis were excluded from this study.

The primary diagnosis was based on both barium enema and full-thickness biopsy results in 20 patients and the findings on barium enema imagingalone in 15 patients. Most of the patients (27/35) had a well-defined transition zone between rectal and descending colonic regions, whereas the remaining patients had no obvious transition zone on imaging.

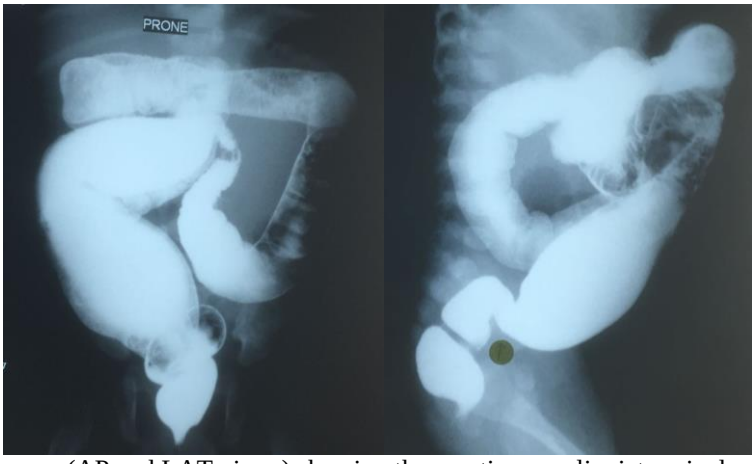

Fig. (1): Barium enema (AP and LAT views) showing the spastic aganglionicterminal rectum, the tapering funnel shaped transition zones and the dilated proximal large bowel in patients with Hirschsprung's disease.

Surgical technique

All patients were admitted to the hospital, 2 - 5 days prior to the surgical intervention and had colonic decompression twice daily. TSPT was considered when the patients had clinically improved or had no impacted faeces. The patients fasted for 6 to 8 hrs.and had rectal irrigation in the morning before surgery. Preoperative antibiotics included intravenous metronidazole and gentamicin.

After having been anesthetized, the infants were placed supine with the pelvis elevated at the edge of operating table and the legs hung down from an inverted U-shaped bar or placed in a lithotomy position in older children. Then patients were prepared circumferentially from the costal margins to the feet. Urinary catheterization was routinely performed. The anal canal was dilated and exposed with Lone Star retractor.

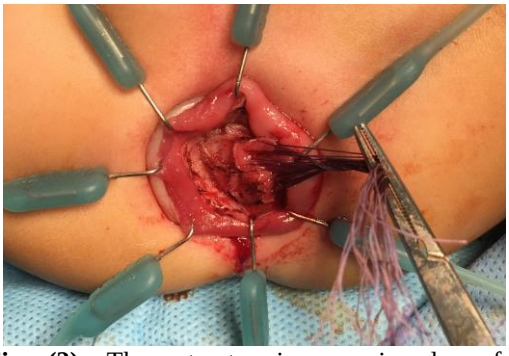

Fig. (2): The retractor is seen in place for adequate visualization and dissection in the pericolic plane.

Transanal rectosigmoidectomy was performed by a circumferential incision using cautery at the level of 1 cm above the dentate line. The entire rectum and part of the sigmoid colon were then delivered through the anusby the use of a circumferential row of 4-0 silk stay sutures placed at the cut end of the colon (just proximal to the incision) in order to facilitate traction during pull-through operation. The full-thickness dissection was then extended superiorly around the rectum by blunt dissection and electrocautery. Extra care was sought during dissection of the retrorectal tissue anteriorly to avoid injuring the urethra or vagina.

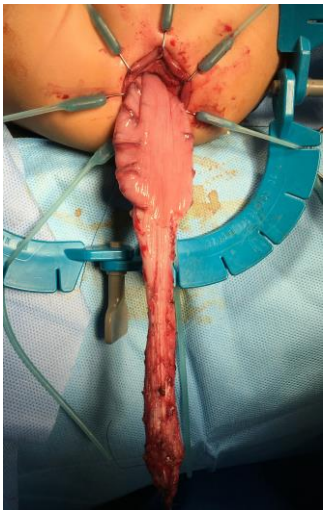

Fig. (3): Rectal pull through the anal orifice after dissection till the transitional zone.

Once the level of peritoneal reflection had been reached, the rectosigmoid was readily mobilized outside the anus. Ligatures were used to dissect rectosigmoid vessels in older children but not in infants. The transition zone was located based on prior biopsy or level corresponding to barium enema findings. The ganglionic proximal colon was anastomosed to the anus using 4-0 interrupted Vicryl sutures.

In the case of abdominal assisted (TSPT) a 2- to 3-cm left lower quadrant incision then was made in a prominent skin crease and the abdomen opened.Seromuscular biopsy specimens were taken from the antimesenteric surface of the colon and sent for frozen section analysis. Once ganglion cells were found, the peritoneal reflection was dissected off of the colon intraabdominally.

Trophic feeding was allowed on the 5th. postoperative days which gradually advanced into an ordinary diet prior to discharge. All patients except two with intraoperative urethral injury could tolerate the routine follow-up at 2 weeks, 1, 3, 6, and 12 months, and are expected to furtherly follow up at a 6 month interval.

RESULTS

Patient demographics, age at diagnosis and at definitive repair, operating time, weight at surgery, length of stay, length of follow-up, and level of the transition zone were documented. Postoperative complications and functional outcome were evaluated.

The series consisted of 22 males and 13 females. There were 8 patients 3 month old, 19 patients between 3 month and 1 year, 5 patients between 1 year and 2 years &3 patients between 2 years and 3 years.

Associated anomalies included Down's syndrome in 2 (5.7%) patients, cardiac anomalies in 4 (11.4%), anorectal malformation in 2 (5.7 %), and cleft lip and cleft palate in 1 (2.8%).

The predominant presenting symptom of Hirschsprung's disease was abdominal distension in 22 of 35 (62.8 %), bilious vomiting in 8 of 35 (22.8%), non-bilious vomiting in 2 of 35 (5.7%), failure to pass meconium in 27 of 35 (77.1%), and constipation in 33 of 35 (94.2%) patients.

According to the radiographic findings, the aganglionic segments were confined to rectum in 5 patients, recto-sigmoid in 27, and descending colon in 3.

We needed to perform abdominal assisted TSPT in 5 patients with ages ranging between 1 to 3 years. This decision was taken prior to the operation based on pre-operative imaging revealing dilated colon that makes it difficult to do all the procedure trans-anally, and off course this added extra minutes to the whole operation but made it easier.

Patient characteristics are summarized in table no (1). Patients' ages at time of operation ranged between 3 months and 3 years with a mean age of 10.4 months +/- 8.33 months. The operative procedure lasted between 90 min and 170 min with a mean of138.14min +/- 26.36 min. There was an apparent difference in operative time between the patients up to 6 month of age (119.1 ± 13.3 min) and patients greater than

6 months (152.3 ± 8.6 min). The extended operative time in older patients was attributed to tedious dissection, not easily controllable blood supply, very thick mesentery, dilated and hypertrophied colon and if added abdominal incision.

The length of resected bowel ranged between13.5 and 27 cm with a mean of 21.05cm±4.41cm. The shortest colonic segment was resected from a 3-month- old boy while the longest colonic segment was resected in a 2.5-year-old boy.All resected segments where sent for pathological examination post-operatively, and all of them confirmed clear margins.

The estimated blood loss was much more in older children compared to the younger infant group, yet not to the extent of needing blood transfusion. Average length of hospital stay was 8.48 ± 1.15 days (range, 7 to 10 days).

Table (1): Collectively showing patient characteristics in the study.

Patient Characteristics	*Min.*	*Maxi.*	*Mean ± SD*
Age at time of surgery	3 months	3 years	10.4 +/- 8.33 mns
Operative time (min)	90 min	170 min	138.14 ± 26.36 min
Length of resected bowel (cm)	13.5 cm	27 cm	21.05 +/- 4.41 cm
Length of hospital stay (days)	7 days	10 days	8.48 ± 1.15 days

Postoperative complications (as collectively demonstrated in table 2) included anastomotic stricture in 2 patients (5.7 %), enterocolitis in 3 patients (8.5 %), urethral injury in 2 patients (5.7 %),Perianal excoriation in 3 patients (8.5 %) and fecal incontinence in 1 patient (2.8 %). The overall complication rate is 11/35 (31.4 %).

We had no anastomotic leaks or bowel obstruction.

Table (2): Collectively demonstrating the complications encountered post operatively.

Complications	*No. of patients (% out of total 35)*
Anastomotic stricture	2 (5.7 %)
Enterocolitis	3 (8.5 %)
Urethral injury	2 (5.7 %)
Perianal excoriation	3 (8.5 %)
Fecal incontinence	1 (2.8 %)
Anastomotic leaks	None
Bowel obstruction	None
Overall complications	11/35 (31.4 %).

Defecation rate of 1–3 times per day was noted in 80% (28/35) of cases within six months of surgery and 14.2% (5/35) at 1 to 2 years post surgery. There were no mortalities in the series.

DISCUSSION

Hirschsprung's disease (HD), or colonic aganglionosis, was first described clinically by Harald Hirschsprung and later precisely defined by Ovar Swenson. Surgical therapy has been the cornerstone of management for the correction of colonic aganglionosis since Swenson described the first successful pull-through operation in 1948.[20]

The TSPT is a recently introduced technique for the treatment of children with HD as an attempt to overcome drawbacks of prior techniques, as in this procedure, the retained aganglionicseromuscular cuff is completely removed. This technique is still not a popular approach, yet attributed to the encouraging results in several studies, we were intrigued and hence came the aim of our study which is to report our experience and compare our initial results to those of others as regards the TSPT surgical procedure.

The average age of patients (included in this study) at the time of surgery is 10.4 months, ranging from 3 months to 3 years. Transanal rectosigmoidectomy was performed by a circumferential incision using cautery at the level of 1 cm above the dentate line, as Paiboon et al,[21] had advocated in his study in 2009. Ho¨llwarth et al.[22] had demonstrated that a low anastomosis at or distal to the dentate line may damage delicate nerve endings that play a role in anorectal continence.

Dissection of the aganglionic segment was pretty stressful as we were aiming at confining the dissection plane as close to the bowel wall as possible, to avoid injuring thepresacral plexus and anterior structure of the pelvis, such as urethra or vas deferens in boys and vagina in girls, but

unfortunately the urethra was injured in two patients.

Another critical issue was related to the evaluation of the transition zone. With an abdominal approach, surgeons may confirm the pathologic results by readily performing multiple biopsies tempted in this technique would considerably affect the operative time. In our study we did abdominal assisted TSPT in 5 patientswith dilated colons in their pre operative enemas, and we found it very beneficial although it prolongs the operative time.Some authors as Georgeson et al. and Langer et al[23,24] had proposed that a laparoscopic biopsy prior to transanal pull-through operation in all cases, where the transition zone is uncertain is helpful to evaluate the real site of transition zone yet we did not do so in our study.

Enterocolitis is considered a potentially lethal complication in patients with HD, occurring either before or after definitive surgery. When compared to a study performed by Sherman et al.[25], with a postoperative enterocolitis rate of 33% and another study advocated by Paiboon et al.[21] with a rate of 11%, we had a considerably low rate of 8.5% in our study.

Anastomotic stricture was another postoperative complication encountered at a rate of 5.7% in our study. In a wide scaled study of 3,618 patients performed by Snyder CL, et al. 2000 who were subjected to various techniques of pull-through for HD, the overall incidence of anastomotic stricture was 7.3% [26].

While Paiboon et al [21] obtained a higher rate of post surgical anastomotic strictures reaching 22.2%. But these strictures were predominantly subclinical strictures detected by routine per rectal examination at the time of follow-up without frank clinical obstruction.

In another study conducted by Ana Christina, et al. [27] attempting to compare results of TSPT technique with those of Duhamel procedure; they had a rate of 22.8% for perineal dermatitis which was much higher than our rate of 8.5% and as for the anastomotic strictures, they were at a rate of 8.6% which is slightly higher than our 5.7%. In this study, results were compared to those of Duhamel procedure revealing no difference in the incidence of postoperative enterocolitis.The incidence of wound infection was lower in the TSPT group. Moreover, the TSPT and Duhamel groups showed no difference in the incidences of mortality, postoperative partial continence, and total incontinence. Yet patients subjected to TSPT had shorter operating times and began oral feeding more quickly after the operation when compared to those subjected to Duhamel procedure.

One patient was complicated with fecal incontinence that warranted intervention. Postoperative encopresis in our study was acceptable for up to 6 months to a year acquiring a regressive course. It was attributed to functional constipation or high amplitude propagating contractions through the neorectumas previously explained by Menezes et al.[28].

Our preliminary results from TSPT are promising in the fact that rate of complete defecation control was as high as 97.2% with the majority accomplished in the first six months'post-surgery.

These results were more or less comparable to those of a study conducted by Kaul A, et al.[29] that included 38 patients who had an average of 0.07 HPACs/min while fasting and 0.13/min in the postprandial state.

We had no anastomotic leaks or bowel obstruction as was the case with the multinational retrospective study performed by Sherman et al.[25], where they came across an anastomotic leak rate of 5.6%, a bowel obstruction rate of 6%, and a considerably high postoperative enterocolitis rate of 33%.

In addition, the average operating time was 4.43 hours in the Sherman et al study[25] which was significantly less in our study being of an average of 2.30 hours, and their average length of stay was 13.6 days which is again longer than the average patient stay in our study that was 8.48 days.

As for the study performed by Bryan C., et al.[30], our results came comparable to theirs in terms of having no anastamotic leaks or bowel obstructions, yet their postoperative enterocolitis rate was 13%; much higher than our 2.8%. Their mean operating time was 2.6 hours which is comparable to our 2.3 hours, yet their mean length of hospital stay was 4 days which is less than our 8.48 days.

Since Swenson and Bill published their description of a rectosigmoidectomy for Hirschsprung's disease in 1948, the traditional approach to the treatment of these patients has been a 2 or 3 stage procedure beginning with a

diverting colostomy.[20] More recently, however, surgeons have started to perform the definitive operation in a one-stage fashion in the infancyperiod. Till now several modifications have been advocated to improve the initial one-stage techniqueincluding selection of level of dissection as regards dentate line and avoiding the deep intra-pelvic dissection thus reducing the risks of sacral nerves as well as the ejaculatory ducts injury, that eventually leads to reduction in hospital stay and morbidity rates.

We were concerned about the fact that as the infant grows, so will any left behindaganglionic bowel that may lead to an increased predisposition to constipation problems, that's why we chose to carry the dissection above the transition zone (by about 6–8 cm) to a level of normal appearing bowel.

Our preliminary results are promising in that they provide further evidence that a one-stage procedure is a safe alternative to the staged procedures. Our technique is feasible, and the early results show it to be safe in all ages with the elimination the need of a colostomy.It is worth nentioning that the abdominal assisted TSPT gave comparable results with only relative prolongation in operative time.

Bryan et al.[30] believe that either a small laparotomy incision or a laparoscopic examination increases the margin of safety by positively identifying the transition zone before beginning the transanal dissection.

The results of more studies are collectively compared to ours, for transanal Swenson's procedure as shown in **[Table 3].**

Series	*No of patients*	*Preop diagnosis*	*Leak*	*Entero-colitis*	*Stricture*	*Dilatation*	*OT (minutes)*	*Blood loss (ml)*	*Urologic Problems*
Gao et al., 2001 [33]	33	Barium enema, manometry & rectal biopsy	None	6.06%	3.03%	Yes	160 (85 – 260)	45 (15 – 100)	NA
Weidner et al., 2003 [34]	15	Rectal biopsy	None	13%	NA (Not Available)	Yes	158 (110 – 190)	NA	NA
Peterlim et al., 2003 [35]	22	Rectal biopsy	9%	Nil	NA	NA	NA	NA	NA
Xu ZL et al., 2008 [36]	134	Barium enema, or rectal biopsy	None	2.9%	NA	NA	70 (50 -115)	NA	NA
Sookpotarom et al., 2009 [37]	27	Barium enema, or rectal biopsy	None	11.1%	22.2%	Yes	153 (60 – 400)	NA	Urethral injury in 1 patient
JK Mahajan et al., 2012 [38]	17	Clinical features & Barium enema	None	11.7%	11.7%	Yes (2 patients)	141 (120 – 200)	58.5 (40 – 180)	None
The current study (our study)	35	Barium enema &/or rectal biopsy	None	8.5%	5.7%	Yes (1 patient)	138 (90 – 170)	55.2 (30 – 110)	Urethral injury in 2 patients

In studies that compared the technical limitations and complications of Soave and Swenson procedures as Leily Mohajerzadeh, et al.[31], they concluded that constipation, cuff abscess and enterocolitis were more common in patients who underwent soave pullthrough surgery attributed to the presence of a retained aganglionic muscular cuff around normal ganglionic colon which represents a portion of retained dysfunctional muscle which is not the case in Swenson pullthrough surgery where the aganglionic segment is resected ending by a colo-anal anastomosis. Otherwise the analyzed 54 patients (Soave 27, Swenson 27) showed no significant differences regarding mean operating time, hospital stay, or fecal incontinence.

From our point of view, the advantages of this one-stage transanal procedure are numerous,

including the avoidance of multiple surgeries and the former prior need for a diverting colostomy with their associated complications, especially with the fact that parents would definitely like to avoid the need to care for a child with a colostomy.

There is also the reduced operating time, less blood loss especially in young infants less than one year of age, reduced risk of pelvic structure damage, single hospital admission, short hospitalization, significantly lower hospital costs, and no external scars with improved cosmetic outcome. Such advantages have been supported by other authors as Paiboon et al., Ana Christina, et al., and Bryan et al.[21,27&30] to avoid the complications of the cuff.

CONCLUSION

In conclusion, our initial results are really promising. Transanal Swenson procedure is an appealing technique overcoming the tedious steps of multistage approaches. It not only avoids multiple abdominal operations, but also the problems associated with the retained dysfunctional muscular cuff of the transanal Soave's procedure. By applying a meticulous dissection strategy, injury to the surrounding structures can be avoided and blood loss can be minimized. So we can conclude that this technique has acceptable short-term outcomes. A long-term follow-up would still be necessary to assess the real impact of the technique on children's wellbeing as they grow up.

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