

One-stage transanal Swenson procedure for rectosigmoid Hirschsprung's disease in infants and children

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Objective This study aimed to present the outcome of transanal one-stage Swenson pull-through procedure in the management of rectosigmoid Hirschsprung's disease (HD).

Background HD is a common cause of intestinal obstruction in pediatric age. Several pull-through procedures have been used to treat this pathology.

Patients and methods Between June 2008 and June 2015, 84 children with biopsy-proven HD underwent transanal one-stage Swenson pull-through procedure. Intraoperative details, postoperative complications, and bowel habits were recorded. Follow-up period ranged from 6 to 42 months.

Results The age at the time of surgery ranged from 3 months to 2 years. The length of the resected aganglionic segment ranged from 12 to 34 cm. The operating time ranged from 72 to 180 min. Postoperative hospital stay ranged from 3 to 6 days. There were no anastomotic leaks, no perianal infection, or postoperative bowel obstruction. Twelve patients (14.28%) developed postoperative enterocolitis. Six patients (7.14%) required a posterior internal sphincter myectomy despite repeated dilatations. All patients had less than four times bowel motions per

day, 3 months after surgery. No voiding disturbances were encountered at the end of the follow-up period and none of the patients complained of recurrent constipation. Six patients developed perianal dermatitis, which was treated conservatively within 3 months after surgery. Anastomotic circumference could not be felt at digital examination in 78 patients 3 months after surgery.

Conclusion One-stage transanal Swenson pull-through procedure is a safe alternative and simpler procedure for rectosigmoid HD with low morbidities and accepted outcome as regards postoperative bowel habits. *Ann Pediatr Surg* 12:104–108 © 2016 Annals of Pediatric Surgery.

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Introduction

Hirschsprung's disease (HD) is the most common cause of intestinal obstruction in newborns. Seventy to 80% of patients have an aganglionic segment at the level of the rectosigmoid colon. Using laxatives or a rectal tube and saline irrigation can successfully treat functional obstruction avoiding colostomy during the neonatal period. The surgical management of the rectosigmoid HD is rapidly changing from the three-stage procedure to a single-stage, transanal, pull-through procedure. This evolution aims at reducing the cost, hospital stay, and the morbidity associated with the staged procedures [1]. Transanal procedures leave no scars, have less postoperative pain, a shorter hospital stay, and lower cost without an increased risk for complications [2,3]. The most commonly used technique for the transanal pull-through procedure is endorectal dissection, which leaves a long muscular cuff that is usually splitted posteriorly. It is well known that the long muscular cuff that is left behind may be a cause of obstruction [4]. A few studies in the literature have shown that the problem of the remnant rectal cuff can be avoided if the transanal resection of the aganglionic segment is performed in the manner described by Swenson – by dissecting the full thickness of the rectum [5,6]. In this study, we reported our experience in transanal Swenson in the last 10 years.

Patients and methods

From June 2008 to June 2015, 84 children diagnosed as having HD, which was confirmed by using barium enema and full thickness rectal biopsy findings, were included in the study. The transitional zone between normal and aganglionic colon was confirmed through intraoperative biopsy analyzed by frozen section. Patients with proven tissue diagnosis of rectosigmoid HD, who were operated as one-stage transanal Swenson procedure, were included in this study. Neonates, older children (> 2 years), patients with either severe enterocolitis or neglected bowel obstruction not responding to bowel decompression, or those who were referred to us after performing initial colostomy, and patients with long aganglionic segments (proximal to sigmoid colon) were excluded from this study.

All children underwent the primary transanal Swenson pull-through procedure. Patient's demographics, age at diagnosis, age at definitive repair, weight at surgery, level of transitional zone, operative time, length of hospital stay, follow-up period, postoperative complications, and functional outcomes were evaluated.

This study was approved by the ethical committee of pediatric surgery department, Ain Shams University.

Preoperative preparation

Preoperative colonic preparation was started 1 day before surgery. The colon was decompressed with saline enemas, and the patient was fed clear fluids only. Six to eight hours before surgery, the patient fasted, and intravenous ampicillin 100 mg/kg/day, gentamycin 2.5 mg/kg/day, and metronidazole 15 mg/kg/day were administered and continued for the first 72 h after the procedure. An informed written consent was taken from all parents or guardians before any surgical intervention.

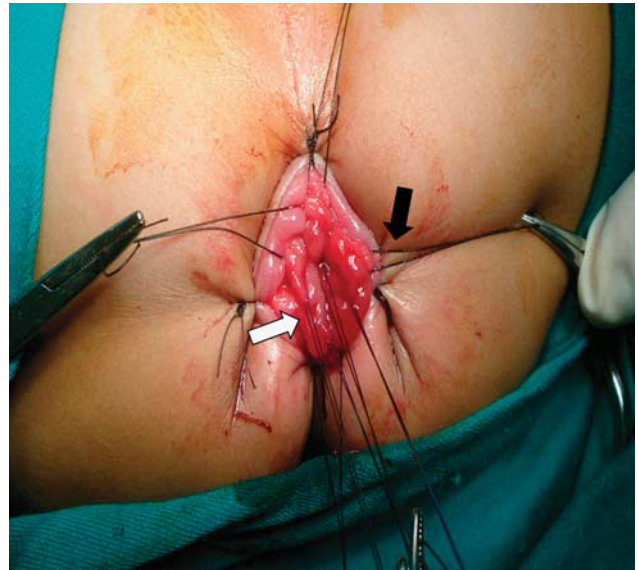
Surgical technique

After the induction of general endotracheal anesthesia, rectal irrigation was performed with a dilute solution of betadine. The patient was then prepared circumferentially from the costal margin to the feet. A Foley's urinary catheter was placed. The anus was gently dilated with Hegar dilators up to 12 or 13 Fr to facilitate the perirectal dissection. Traction sutures were placed just proximal to the anoderm but distal to the dentate line in a circumferential fashion and secured to the buttocks, thus everting the anus. A circumferential row of 4-0 silk stay sutures was inserted ~0.5–1 cm above the dentate line. A full thickness circumferential incision was made just above the circumferential silk stay sutures by using a fine diathermy needle. Stay sutures (4-0 vicryl) were placed in the cut end of the colon to assist with traction (Fig. 1), and then the dissection was carried proximally. Care was taken to keep the dissection on the surface of the serosa, without straying laterally (Fig. 2). Vessels were controlled either with the monopolar electrocautery or tied (Fig. 3). The dissection was continued up to the level where transitional zone could be seen. During this step, full thickness biopsy specimens of the colon were examined by frozen section to assure normoganglionic level. After frozen section confirmation of ganglion cells was reported, the colon was freed without tension up to the proposed anastomotic line (Fig. 3); a colectomy of the dilated and thickened ganglionic segment was performed. A single-layered, full thickness anastomosis was created with 4-0 interrupted, absorbable sutures [7].

Postoperatively, oral feeding was started with the resumption of gut function. Discharge occurred when pain was controlled on oral medications, and the patient was tolerating a regular diet appropriate for age. The patient was examined in the clinic 2 weeks later where the anus was sized with Hegar dilators. Routinely, the parents were taught to perform anal dilatations, gradually increasing the size of the dilator by 1 U every 15 days and stopped on the basis of the child's age as suggested by Pena for children with anorectal malformations [8].

Patients were followed up every 15 days for the first 2 months, on a monthly basis for 3 months, and then every 2 months thereafter. Patients with more than eight bowel movements per day were considered as having diarrhea. Patients requiring enema or medical regimen or both to have regular bowel movements were considered as having constipation. Anastomotic stricture was diagnosed if more than home dilatations was required. Enterocolitis was

Fig. 1



Above dentate line traction sutures (black arrow), proximal vicryl stay sutures (white arrow).

Fig. 2



Dissection on the colon just at the surface of the serosa.

defined as the clinical syndrome characterized by abdominal distension, diarrhea and fever above 38°C.

Results

Eighty-four patients with rectosigmoid HD were surgically managed by one-stage transanal pull-through Swenson procedure. There were 54 boys (64.28%) and 30 girls (35.72%) in this series. The median age at diagnosis was 17 days, ranging from 3 days to 20 months. The median age at surgery was 11 months, ranging from 3 months to 2 years. The median weight at the time of surgery was 5.125 kg, ranging from 3.5 to 12 kg. The median length of aganglionic bowel resected transanally was 22.5 cm, ranging from 12 to 34 cm. The operative time ranged between 72 and 180 min (median of 85 min). No patient required conversion to an abdominal procedure. There were no intraoperative complications. There was minimal blood loss with no patient requiring a blood transfusion. There were no anastomotic leaks, perianal infection, or bowel obstruction. Bowel movements

Fig. 3



Ligation and electrocautery of rectosigmoid mesenteric vessels (white arrow) during colon mobilization; macroscopic transitional zone can be observed (black arrow).

returned to normal within 24 h after surgery in all patients. Progression in oral intake of feedings was uneventful, and full oral intake was achieved on the third to the fifth postoperative day. The median postoperative hospital stay was 5 days, ranging from 3 to 6 days. The median follow-up period was 9 months, ranging from 6 to 42 months. All patients underwent initial postoperative anal dilatations; 78 out of 84 patients did not require long-term dilatations.

Twelve patients (14.28%) suffered attacks of postoperative enterocolitis, which were treated successfully with intravenous metronidazole, cephalosporin, and rectal irrigations. Six patients (7.14%) required a posterior internal sphincter myectomy secondary to poor emptying because of the presence of anastomotic stricture and rectal stenosis despite dilatations. In 48 patients (57.14%), the maximum number of defecations were less than four times per day after the definitive surgery. Frequent bowel movements were observed in 36 patients (42.85%); in these patients the number of defecations was greater than eight times per day and soiling three to four times per day. The number of bowel movements decreased to three to four times per day and soiling one to two times per day for these 36 patients at the end of 3 months postoperatively. No voiding disturbances in the form of incontinence, poor stream, or retention were encountered at the end of the follow-up period and none of the patients complained of recurrent constipation. These 36 patients also had different grades of perianal

dermatitis caused by frequent stools and soiling. Perianal dermatitis was effectively treated with topical zinc-based barrier cream and decrease in the number of stools. At digital examination of 78 patients (92.86%) at 3 months after surgery, the anastomotic circumference could not be felt.

Discussion

In the last decades, surgeons have started to perform the definitive operation for HD in a one-stage fashion even in the neonatal period, which has been shown to be successful in many open series using a Soave, Duhamel, or Swenson procedures [9–13]. Smith *et al.* [14], Curran and Raffensperger [15], and Georgeson *et al.* [16] described different primary laparoscopic pull-through procedures for HD in infants and children. This minimally invasive laparoscopic technology has improved many aspects of the surgical treatment of HD, but this technique still requires laparoscopic dissection of the rectum with the associated risk for bleeding and thermal or harmonic injury to other pelvic organs [17]. The completely transanal approach offers same advantages as laparoscopic surgery (reduced or no postoperative ileus, less postoperative pain, and early hospital discharge), but carries with it added advantages: elimination of risks associated with intra-abdominopelvic dissection such as bleeding, injury to other organs, adhesion formation, less postoperative pain from the absence of multiple abdominal port sites, better cosmetic results, and reduced costs when compared with the laparoscopic technique [18,19].

In this series, one-stage Swenson pull-through procedure was performed totally transanally, thus avoiding the deep intrapelvic dissection, which was adopted by Bryan and John [7], as they started the procedure by committing minilaparotomy in the left lower quadrant of the abdomen to dissect and skeletonize the distal normoganglionic colon down to the perineal reflection. By keeping the dissection directly on the bowel wall, and by using urethral catheter, risk to the sacral nerves as well as the ejaculatory ducts in boys are minimized. In addition, this procedure decreases the risk for bleeding, cuff abscess, and postoperative constipation, which can occur in patients who had been subjected to transanal endorectal pull-through procedure because of the presence of the muscle cuff of the distal aganglionic segment [1,20,21].

One of the important factors, particularly in infants and young children, is where to begin and how far proximal to carry the dissection. In the current series, the dissection started 0.5–1 cm above the dentate line, thus removing all aganglionic bowel. Our concern was that as the infant grows, the aganglionic segment may lengthen over time, and there may be an increased predisposition to constipation. Second, the dissection above the transitional zone to a level of normal-appearing bowel was chosen. In several children, the ganglionated bowel was dilated to such an extent that resection of the dilated segment was warranted to allow for a better coloanal anastomosis, and to avoid potential motility problems with the dilated segment.

The median operative time in this series was 85 min. Omitting the submucosal dissection in the Swenson procedure may explain the less time taken to complete the procedure, which is less than that reported by Sherman *et al.* [22], George *et al.* [23], Sumate [24], and Mahajan *et al.* [25], who had an operative time of 4.4 h, and 150, 96, and 141.7 min, respectively, but more than that in a study by Zhi-lin *et al.* [26] who recorded an average operative time of 70 min.

In a review of 880 Swenson procedures (a combination of two-stage or three-stage), Sherman *et al.* [22] reported an anastomotic leak rate of 5.6%. In another series by Hadidi [27], anastomotic leak rate of 3% was reported, which may be attributed to coloanal anastomosis being fashioned under some degree of tension, or to ischemia. In the current series, there was no anastomotic leak, which is in line with that findings in a study by Dela Torre-Mondragon and Ortega-Salgado [18], George *et al.* [23], and Orkan *et al.* [28].

In this series, there was no postoperative adhesive intestinal obstruction. It is well-known that minimally invasive surgery reduces the incidence of intra-abdominal adhesions [20]. This is probably more true for the patients for whom a purely transanal approach is used, in which the incidence of this complication should approach zero. In contrast, the reported incidence of adhesive small bowel obstruction after open pull-through procedure for HD has ranged from 2 to 20% [22,29,30].

Enterocolitis was noted in 12 patients (14.28%), which is in agreement with the reported incidence of postoperative enterocolitis in other series, ranging from 10 to 33% [29, 31–34]. In 84 patients with HD treated by So *et al.* [21], the authors did not encounter postoperative enterocolitis or stricture and attributed this in part to early and adequate dilatations. Hackman *et al.* [35] studied the risk factors for postoperative enterocolitis and found that both the presence of anastomotic leak or stricture and the development of postoperative intestinal obstruction secondary to adhesions increased the relative risk and subsequent enterocolitis by approximately three-fold. These risk factors increase intestinal stasis and create the cycle (stasis–bacterial overgrowth–mucosal invasion) leading to the subsequent local and systemic inflammatory response [36]. The relative low incidence of enterocolitis after one-stage transanal Swenson procedure in the current series may be related in part to the absence of seromuscular cuff, the low coloanal anastomosis, and the policy of routine postoperative anal dilatation. In the current study, anastomotic stricture and rectal stenosis occurred in six patients (7.14%). The rate of these complications has been reported as 15.7–22% [32], although this rate is less than that reported by Mahajan *et al.* [25] (11.7%), but still higher than that reported by Umar *et al.* [34] (4%).

When the transanal endorectal pull-through was introduced, some authors reported less continence capacity compared with the classic transabdominal approaches [36]. The initial argument was that the overstretching of the anal sphincter, during the transanal

operation, could be a critical issue affecting continence. To address this matter, several studies have been published and, in particular, Kim *et al.* [37], examined long-term stooling outcomes in a large, multicenter cohort of patients undergoing either transanal endorectal pull-through or the transabdominal approaches. Transanal endorectal pull-through procedure was associated with fewer complications, fewer episodes of enterocolitis, and no higher incidence of incontinence [18,38,39]. In the current study, there was no stooling disturbance at the end of the follow-up period and this can be explained by the shorter length of operation, applying just the right amount of dilatation, with no overstretching of the anal sphincter.

Conclusion

Transanal Swenson is feasible, and avoids the problems associated with the long muscular cuff of the transanal Soave's procedure with excellent cosmetic results. The modification of the Swenson procedure to a transanal dissection as opposed to an intrapelvic dissection has the additional potential advantage of avoiding injury to intrapelvic structures, preserving the sphincters, blood supply and innervation; therefore, urinary and fecal continence are less likely to be offended.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

References

- Höllwarth ME, Rivosecchi M, Schlee J, Deluggi S, Fasching G, Ceriati E, *et al.* The role of transanal endorectal pull-through in the treatment of Hirschsprung's disease – a multicenter experience. *Pediatr Surg Int* 2002; **18**:344–348.
- Albanese CT, Jennings RW, Smith B, Harrison MR. Perineal one-stage pull-through for Hirschsprung's disease. *J Pediatr Surg* 1999; **34**:377–380.
- Langer JC, Seifert M, Minkes RK. One-stage soave pull-through for Hirschsprung's disease: a comparison of the transanal and open approaches. *J Pediatr Surg* 2000; **35**:820–822.
- Rintala RJ. Transanal coloanal pullthrough with a short muscular cuff for classic Hirschsprung's disease. *Eur J Pediatr Surg* 2003; **13**: 181–186.
- Weidner BC, Waldhausen JH. Swenson revisited: a one stage transanal pullthrough procedure for Hirschsprung's disease. *J Pediatr Surg* 2003; **38**:1208–1211.
- Peterlim FL, Martins JL. Modified transanal recto-sigmoidectomy for Hirschsprung's disease: clinical and manometric results in the initial 20 cases. *J Pediatr Surg* 2003; **38**:1048–1050.
- Bryan CW, John HTW. Swenson revisited: a one-stage, transanal pullthrough procedure for Hirschsprung's disease. *J Pediatr Surg* 2003; **38**:1208–1211.
- Pena A. Current management of anorectal anomalies. *Surg Clin North Am* 1992; **72**:1393–1416.
- Cass DT. Neonatal one-stage repair of Hirschsprung's disease. *Pediatr Surg Int* 1990; **5**:341–346.
- Cilley RE, Statter MB, Hirschl RB, Coran AG. Definitive treatment of Hirschsprung's disease in newborn with a one-stage procedure. *Surgery* 1994; **115**:551–556.
- Pierro A, Fassoli L, Kiely EM, Drake D, Spitz L. Staged pull-through for rectosigmoid Hirschsprung's disease is not safer than primary pull-through. *J Pediatr Surg* 1997; **32**:505–509.
- Santos MC, Giacomantonio JM, Lau HYC. Primary Swenson pull-through compared with multiple stage pull-through in the neonate. *J Pediatr Surg* 1999; **34**:1079–1081.
- Wilcox DT, Bruce J, Bowen J, Bianchi A. One-stage neonatal pull-through to treat Hirschsprung's disease. *J Pediatr Surg* 1997; **32**:243–247.
- Smith BM, Steiner RB, Lobe TE. Laparoscopic Duhamel pull-through procedure for Hirschsprung's disease in childhood. *J Laparoendosc Surg* 1994; **4**:273–276.
- Curran JJ, Raffensperger JC. The feasibility of laparoscopic Swenson pull-through. *J Pediatr Surg* 1994; **29**:1273–1275.

- 16 Georgeson KE, Fuenfer MM, Hardin WD. Primary laparoscopic pull-through for Hirschsprung's disease in infants and children. *J Pediatr Surg* 1995; **30**:1017-1022.
- 17 Langer JC, Minkes RK, Mazziotti MV, Skinner MA, Winthrop AL. Transanal one-stage Soave procedure for infants with Hirschsprung's disease. *J Pediatr Surg* 1999; **34**:148-152.
- 18 Dela Torre-Mondragon L, Ortega-Salgado JA. Transanal endorectal pull-through for Hirschsprung's disease. *J Pediatr Surg* 1998; **33**:1283-1286.
- 19 Wester T, Rintala RJ. Early outcome of transanal endorectal pull-through with a short muscle cuff during the neonatal period. *J Pediatr Surg* 2004; **39**:157-160.
- 20 Garrad CL, Clements RH, Nanney L, Davidson JM, Richards WO. Adhesive formation is reduced after laparoscopic surgery. *Surg Endosc* 1999; **13**: 10-13.
- 21 So HB, Becker JM, Shawartz DL, Kutin ND. Eighteen years experience with neonatal Hirschsprung's disease treated by endorectal pull-through without colostomy. *J Pediatr Surg* 1998; **33**:673-675.
- 22 Sherman JO, Snyder ME, Weitman JJ, Jona JZ, Gillis DA, O'Donnell B, et al. A 40-year multinational retrospective study of 880 Swenson procedures. *J Pediatr Surg* 1989; **24**:833-838.
- 23 George E, Diego F, Fabio T, Merulla VE, Manciana A, Caccia G. Further evidence on totally transanal one-stage pull-through procedure for Hirschsprung's disease. *J Pediatr Surg* 2003; **38**:1434-1439.
- 24 Sumate T. Transanal one-stage endorectal pull-through for Hirschsprung's disease in infants and children. *J Pediatr Surg* 2003; **38**:184-187.
- 25 Mahajan JK, Rathod KK, Bawa M, Narasimhan KL. Transanal Swenson's operation for rectosigmoid Hirschsprung's disease. *Afr J Paediatr Surg* 2011; **8**:301-305.
- 26 Zhi-lin X, Zheng Z, Long W, An Q, Tao WF. A new modification of transanal Swenson pull-through procedure for Hirschsprung's disease. *Chin Med* 2008; **121**:2420-2423.
- 27 Hadidi A. Transanal endorectal pull-through for Hirschsprung's disease: experience in 68 patients. *J Pediatr Surg* 2003; **38**:1337-1340.
- 28 Orkan E, Ahmet C, Zafer D, Balik E. Submucosal pressure-Air insufflation facilitates endorectal mucosectomy in transanal endorectal pull-through procedure in patients with Hirschsprung's disease. *J Pediatr Surg* 2003; **38**:188-190.
- 29 Jona JZ, Cohen RD, Georgeson KE, Rothenberg SS. Laparoscopic pull-through procedure for Hirschsprung's disease. *Semin Pediatr Surg* 1998; **7**:228-231.
- 30 Moore SW, Albertyn R, Cywes S. Clinical outcome and long-term quality of life after surgical correction of Hirschsprung's disease. *J Pediatr Surg* 1996; **31**:1496-1502.
- 31 Marty TL, Seo T, Matlak ME, Sullivan JJ, Black RE, Johnson DG. Gastrointestinal function after correction of Hirschsprung's disease: long-term follow up in 135 patients. *J Pediatr Surg* 1995; **30**:655-658.
- 32 Minford JL, Ram RR, Turnock RR, Lamont GL, Kenny SE, Rintala RJ, et al. Comparison of functional outcomes of Duhamel and transanal endorectal coloanal anastomosis for Hirschsprung's disease. *J Pediatr Surg* 2004; **39**:161-165.
- 33 Sharsgard ED, Superina RA, Shandling B. Initial experience with one-stage endorectal pull-through procedures for Hirschsprung's disease. *Pediatr Surg Int* 1996; **11**:480-482.
- 34 Umar FA, Muhammad I, Naz A. Congenital aganglionosis; single stage transanal pullthrough, a big relief to patients. *Professional Med J* 2014; **21**:312-315.
- 35 Hackman DJ, Filler RM, Pearl RH. Enterocolitis after the surgical treatment of Hirschsprung's disease: risk factors and financial impact. *J Pediatr Surg* 1998; **33**:830-833.
- 36 Teitelbaum DH, Cilley RE, Sherman NJ, Bliss D, Uitvlugt ND, Renaud EJ, et al. A decade of experience with the primary pull-through for Hirschsprung's disease in the newborn period: a multicenter analysis of outcome. *Ann Surg* 2000; **232**:372-380.
- 37 Kim AC, Langer JC, Pastor AC, Zhang L, Sloots CE, Hamilton NA, et al. Endorectal pullthrough for Hirschsprung's disease-a multicenter, long-term comparison of results: Transanal vs transabdominal approach. *J Pediatr Surg* 2010; **45**:1213-1220.
- 38 Stefano G, Pietro B, Alessandra N, Grandi F, Midrio P, Mognato G, et al. Outcome comparison among laparoscopic Duhamel, laparotomic Duhamel, and transanal endorectal pull-through: a single-center, 18-year experience. *J Laparoendosc Adv Surg Tech A* 2011; **21**:859-863.
- 39 Stensrud KJ, Emblem R, Bjørnland K. Functional outcome after operation for Hirschsprung disease: transanal vs transabdominal approach. *J Pediatr Surg* 2010; **45**:1640-1644.