Original Article ISSN (O):2395-2822; ISSN (P):2395-2814



# Primary Transanal Swenson's Pull-through in Hirschsprung's Disease in SRHF, Mizoram, India.

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Received: October 2018
Accepted: December 2018



## ABSTRACT

**Background:** The purpose of this study was to analyze the patients of Hirschsprung's disease (HD) who undergone primary Transanal Swenson's Pullthrough operation, its short and intermediate term outcome in neonates, infants and children in the department of General/Pediatric Surgery in State Referral Hospital, Falkawn (SRHF)/MIMER, Mizoram, India. **Methods:** Twenty four patients with rectosigmoid HD underwent single-stage transanal Swenson's procedure. The contrast enema finding with definite transition zone was relied upon for diagnosis. Full thickness rectal dissection was done starting from 0.5-1 cm above the dentate line. The mobilized colon was resected about 5 cm or more above the transition zone. Full thickness colo-anal anastomosis was then performed. **Results:** There were 21 male and 3 female patients and the ages of the patients ranged from 4 days to 3 years. The mean length of the resected colon was 19.54± 9.85 cm. The anatomical transition zone correlated with the pathological transition zone in all the cases. The mean follow up period was 8.28±3.9 months. Two patients had post-operative enterocolitis, and one patient had stricture of the anastomosis. Two patients expired during the follow up period, one due to sepsis and the other due to community acquired pneumonia. One patient continued to have occasional fecal soiling and one patient developed perianal fistula for which diverting colostomy was done. Two patients had ongoing occasional constipation. None of the patients had voiding disturbances or incontinence. **Conclusion:** Primary transanal Swenson's pull through is a safe and viable alternative technique for patients with rectosigmoid HD. The procedure is feasible even in neonates and in upper sigmoid colon HD.

***Keywords:*** Hirschsprung's disease, Swenson, Transanal, Pullthrough, Enterocolitis.

## INTRODUCTION

Following Dr. Orvar Swenson's description of the operative approach to the management of Hirschsprung's disease in 1948, other surgical procedures developed subsequently, including the endorectal dissection (Soave), retrorectal procedure (Duhamel) and a low anterior resection (Rehbein).[1-4] In addition, the last one and half decade witnessed the emergence of the transanal technique, and the addition of laparoscopy to all these procedures.[5-7] The most commonly performed transanal pull-through technique; iethe endorectal dissection has become widely accepted all over the world and had established itself as the primary procedure for most rectosigmoid HD.[8] From a social and economic perspective, the single staged primary transanal procedure is undoubtedly beneficial especially in the developing and poorer countries. It has the potential advantages of reducing the cost, hospital stay, and also morbidity associated with the staged procedures. The primary concern in the endorectal procedure though, is the long muscular cuff that is left behind. This remnant cuff has been implicated for recurrent obstructive symptoms; manifested by recurrent enterocolitis, severe constipation, and overflow incontinence.[9,10] To avoid this cuff related problems, various modifications have tried including a shorter muscle cuff, internal anal sphincterotomy, and oblique anastomosis.[11] The original Swenson's operation was perceived to have a high incidence of complications including urinary incontinence, fecal incontinence, and impotence.[12] However, the full-thickness rectal dissection, if done correctly in the proper plane previous studies have shown that the incidence of the aforementioned complications were found to be less and the results were found to be as good as the other pull-through procedures. It also avoids leaving behind any significant residual aganglionic bowel in the form of a cuff or a pouch.[13-14] In this study, we present our results following the primary transanal Swenson's procedure after adequate diagnosis has been inferred from the preoperative Contrast enema study which was validated by the intraoperative findings and confirmed by the post-operative specimen histopathology and histochemistry.

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## MATERIALS AND METHODS

This study was carried out in the department of Surgery (Pediatric Surgery Unit) in the State Referral Hospital, Falkawn (SRHF)/MIMER (Mizoram Institute of Medical Education and Research), Mizoram, India, during the period of 2 years from July 2016 to June 2018. A total of 24 patients with recto-sigmoid Hirschsprung's disease were selected to undergo the transanal single-stage Swenson's procedure. The study was approved by our institution's Ethics Review Board. Patients were included irrespective of the age group whose contrast radiography showed a definitive recto-sigmoid HD. Exclusion criteria were Patients on colostomy or requiring colostomy due to poor general condition or medical illnesses or not decompressing well by conventional means, doubtful diagnosis by CE or long segment aganglionosis. The presence of enterocolitis was another exclusion criteria and none of the cases required preoperative rectal biopsy for the diagnosis. Preoperative bowel preparation was done with saline rectal wash outs, and intravenous antibiotics were given before the operation. Under general anesthesia and caudal block, a per-urethral catheter of appropriate size was inserted. The operation was performed with the patient positioned in lithotomy position in all except in 2 patients where it was done in the prone position. Full thickness interrupted circumferential 6-8 stay sutures were placed just above the dentate line for traction and eversion of the rectum. The rectal mucosa was incised by monopolar electrocautery just proximal to the traction sutures 0.5 to 1 cm above the dentate line. The incision was deepened to include full-thickness rectal wall and rectal mobilization was done by working on the surface of the rectal wall using a bipolar cauterization probe. The dissection was carried into the peritoneal cavity and the proximal dissection was continued till the transition zone was clearly visible. The normal dilated colon was resected 4-5cm above the transition zone or further more proximally in cases of significant dilated colon. This was done in order to accommodate the normal pulled down colon and facilitate proper colo-anal anastomosis. Intra-operative identification of the anatomical transition zone was possible in all the case. Apart from the conventional H&E (Hematoxylin and Eosin) stain to determine the presence or absence of ganglion cells and hypertrophied nerve fibres, each specimen were examined by immunohistochemistry using a Calretinin stain. The histopathological and immunohistochemical examinations of the aganglionic (narrow), transition zone and normal (dilated) part of the colon correlated with the preoperative CE and anatomical findings in all the patients. Patient's hospital courses and follow-up at 2 weeks, 1 month, 3 months, 6 months and then at 12 months were evaluated. Results were expressed as mean and Standard Deviation (SD) as well as median (IQR- Inter Quartile Range), using one sample t test. We used GraphPadInStat version 3 (GraphPad Software Inc, SanDiego 92130, USA) and Inter-Quartile Range calculator (www.alcula.com) for data analysis.

## RESULTS

After employing our exclusion criteria, 24 patients were selected for the study, with age at the time of operation ranging from 4 days to 3 years with a median age of 112 days (IQR 19-262.5). There were 21 male and 3 female patients with a ratio of 8:1. None of the cases had any associated congenital anomaly. Preoperative contrast enema revealed a transition zone in the rectosigmoid in 20 patients (83.3%), rectum in 2 (8.3%) and upper sigmoid colon in 2 (8.3%).The time from diagnosis to definitive surgery was a median of 6.5 days (IQR 3.25-8.75). The mean duration of operation was 55.6± 16.83 min. (range of 45-90 minutes).The length of the aganglionic segment ranged from 2-45 cm with a mean of 11.8 ± 10.9 cm. The length of colon resection ranged from 8-47 cm with a mean of 19.54± 9.85 cm. With the intraoperative blood loss between 5-30 ml (mean 14.58±6.24 ml), none of the patients were given blood intra-operatively. Except for 1 patient who had an inadvertent vaginal injury which was repaired primarily, the were no intra-operative complications encountered. Apart from the initial dissection requiring more tedious effort in older children due to the presence of well-developed appendices epiploicae and fat on the serous layer of the colon, there were no major difficulty faced during operation. All the 24 patients tolerated oral feeds by the third postoperative day with a median time of 24 hours (IQR 24-24). In all the patients, the first passage of stools was observed in 1-2 days (median 1 day/ IQR 1-1).The hospital stay ranged from 5-15 days (median duration of 5 days/ IQR 5-6.75). Immediate postoperative complications were evaluated. One patient who had intraoperative vaginal injury was given analgesics for 5 days. In the rest of the patients, analgesic was withdrawn on the 2nd day. Intravenous antibiotics were given for 5 days in all except in one patient who developed sepsis on the 5th post-operative day where antibiotic was upgraded. During the early post-operative period, abdominal distension was the most frequent complaint after the operation. Eight patients (33.3%) had abdominal distension, out of which the distension was transient in 4 patients resolving within 2 weeks. The remaining 4 patients had ongoing occasional abdominal distension and were associated with anastomotic narrowing. Four patients had perianal excoriation but all healed by 2 months with local application of barrier creams and perineal care. One patient developed post-operative

enterocolitis, which resolved with antibiotics and rectal wash outs. One neonate who had a poor post-operative recovery following the pull-through operation eventually expired at the 5th postoperative day due to overwhelming sepsis. The patients were followed-up regularly in the out-patient clinic. Those who missed the follow-up schedule were contacted by telephone. Out of the 24 patients included in the study, 14 patients were followed up to 1 year. The mean follow-up period was 8.28±3.9 months [Table 1].

**Table 1: Complications at 12 months.**

| Complications | By 12 months | percentage |
|---|---|---|
| Abdominal distension | 4 (occ.) | 16.6% |
| Anastomotic narrowing | 3 | 12.5% |
| Anastomotic stricture | 1 | 4.1% |
| Enterocolitis | 2 | 8.3% |
| Perianal fistula | 1 | 4.1% |
| Mucosal prolapse | 0 | |
| Constipation | 2 | 8.3% |
| Soiling | 1 | 4.1% |
| Intestinal obstruction | 0 | |
| Death | 2 | 8.3% |

The 4 patients with anastomotic narrowing were managed with regular anal dilatation at home. One patient with poor compliance eventually developed anastomotic stricture and enterocolitis at the completion of 1 year follow up. The patient recovered with an aggressive regimen of antibiotics, rectal wash outs and anal dilatation without the requirement for a secondary operation. One patient developed perianal fistula at months requiring a diverting colostomy. One patient who was lost to the 3rd month follow up was discovered that he had expired at home due to respiratory tract infection. The frequency of stools per day gradually reduced from an average of 10 times per day at 2 weeks to an average of 2-3 times per day at 1 year follow-up. Minor degree of soiling was seen in 2 patients at 6 months follow-up which reduced to 1 patient at 1 year follow-up. The remaining had a dry perineum in between normal bowel movements. By the end of the study, 75% of the patients had normal bowel habits and 87.5% were continent with only 1 patient having occasional fecal soiling. Two patients continued to have ongoing occasional constipation and mild abdominal distension [Table 2].

**Table 2: Functional results**

| Function | No of patients (n=22 at end of the study) |
|---|---|
| Normal bowel habit | 18 |
| Soiling | 1 |
| Constipation | 2 |
| On Colostomy | 1 |

These patients were managed with laxatives, dietary modifications and regular anal bougienage. Urinary continence was noted in all the patients in our series who were old enough to be assessed, and age related milestones for both urinary and fecal continence were not delayed. On further questioning, majority of the caregivers of male patients noticed spontaneous erection during voiding.

**Table 3: Comparison with some published series of TEPT *(yrs- years)**

| Series | No of pts. | Age at operation | Perianal excoriation (%) | Enterocolitis (%) | Stricture/ stenosis (%) | Anastomotic dehiscence (%) | Bowel function |
|---|---|---|---|---|---|---|---|
| Elhalaby et al1995 | 149 | 8D-14yrs | 32.2 | 17.5 | 4.7 | 1(0.7)/cuff abscess 2% | 83.3% continent |
| Shankar et al 2000 | 136 | 1 M | | 10 | 4 | | 76% continent |
| Langer et al. 2003 | 141 | 146 D | 11 | 6 | 4 | 0 | 81% N<br>18% minor dysfunction<br>1% major dysfunction |
| Zhang et al 2006 | 58 | 12M13yrs | 3 | 5 | 0 | | 46% satisfactory |
| Li et al. 2006 | 112 | | 0 | 21 | 11 | 0 | 90% satisfactory |
| Obermayr et al. 2009 | 25 | Mean 3.5M | 0 | 2 | 1 | 0 | 95% continent |
| Van de Ven et al. 2013 | 21 | 2.4 M (0.7-31.6) | 0 | 24 | 0 | 0 | 48% satisfactory |
| Present series 2016 | 24 | 112 days (median)/(4 days-3 yrs) | 16.6 | 8.3 | 4.1 | 4.1 (perianal fistula) | 87.5% satisfactory<br>75% normal habit |

**Table 4: Comparison with various series of Transanal Swenson's procedure**

| Series | No. of patients | Follow up duration (M-months) | Operating time (minutes) | Length of bowel resection (cm) | Blood loss (ml) | Stricture (%) | Leak (%) | EC (%) | Bowel function |
|---|---|---|---|---|---|---|---|---|---|
| Gaoet al,2001 | 33 | 6-18 Median10.5 | 160 (85-260) | 29.5 | 45 | 3.03 | Nil | 6.06 | 84% Normal |

| | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | | habit |
| Weidner et al.,2003 | 15 | 9 (0.5-36) | 158(110-190) | NA | NA | NA | None | 13 | 80% Normal habit |
| Peterliniet al.,2003 | 20 | 29-34 | NA | NA | NA | NA | 9% | Nil | 100% Normal |
| Sookpotaromet al.,2009 | 27 | 12-24 | 153.5±85.9 | 16.3±4.7 | NA | 22.2% | None | 11.1 | 77.8% Normal |
| Mahajan et al., 2010 | 17 | 35.4(6-45) | 141 (120-200) | 18.2(15-29) | 58.5 (40-180) | 11.7 | None | 11.7 | 85% Normal |
| Present series 2016 | 24 | 8.28±3.9M | 55.6± 16.83 | 19.54± 9.85 | 14.58±6.24 | 4.1 | 4.1 | 8.3 | 75% Normal |

## DISCUSSION

The original operative procedure for Hirschsprung's disease, described by Drs. Swenson and Bill in 1948, was a transabdominal approach where careful extra-rectal dissection was carried down to a level two centimeters above the anal canal.[1] It was thought to have a high incidence of complications including urinary incontinence, fecal incontinence, and impotence. The cause was implicated to be a too wide dissection around the rectum leading to injury to the nerviergentes.[12] Other alternative procedures soon followed accordingly. However, a careful review of long-term data appeared to suggest that Swenson's original procedure compares very favorably to other operative techniques.[15] In a manuscript by Sherman et al.1989, describing the outcomes of 880 Swenson procedures, they reported no complications of urinary or sexual problems post-operatively and the rates of leak, reoperation, and postoperative enterocolitis were lower than historical data of other resection techniques.[13] The first report on the transanal Soave procedure for the classic recto-sigmoid HD was published by De La Torre et al in 19988. In addition to minimizing the rate of complications due to laparotomy or the presence of a stoma and decreasing the number of hospitalizations and cost, the avoidance of a colostomy has dramatically improved the quality of care to children with Hirschsprung's disease. The main problem with the endorectal dissection technique is that it leaves a long muscular cuff, which is usually split posteriorly. Proponents of the Soave procedure have suggested various modifications including limiting the amount of residual aganglionic segment (the cuff) to 1-2 cm from the beginning of the dissection, the extent of which has become more Swenson-like, and some authors actually called it a "Soaveson".[16] We have performed the primary Swenson's procedure using the transanal approach in selected cases of Hirschsprung's disease where the preoperative contrast enema showed a definite transition zone in the rectosigmoid region. We did the full-thickness, extrarectal dissection using a bipolar cautery adhering to the principle of staying on the rectal wall. Employing the bipolar cautery also helped us in minimizing spreading electric current and heat to prevent injury to the surrounding nerves and structures. We resected an additional length of a minimum of 4-5cm or more above the transition zone, and in some cases significant length of normal dilated colon was resected to accommodate the pulled down colon for proper colo-anal anastomosis.

The percentage of neonates in this series was 29.1%. In more than one way, this has proved to be advantageous including parental acceptance and technically being easier dissection in neonates. Zhang et al. noted that younger patients and shorter aganglionic segments were associated with better clinical outcomes in TEPT procedure.[17] We also found that the rectal dissection was relatively more challenging in older children (>3 years group) due to the thickness of the mesentery and presence of fat and appendices epiploicae on the gut wall, but we did not encounter any major difficulty during the procedure. With a sensitivity of 65-80% and specificity of 66-100% in literature18, we have employed contrast enema to see the presence of transition zone (TZ), irregular contractions or an abnormal recto-sigmoid index. The pre-operative contrast enema studies in our series were in concordance with the intraoperative findings as well as with the histopathological and immunohistochemical results in all the cases. We did not utilize intra-operative frozen biopsy for any of the cases. Our data substantiated few previous reports that contrast enema is sufficient for the diagnosis of HD and identification of a

Well-defined transition zone in a preoperative contrast enema is enough to perform a pull-through procedure in HD.[19,20]

Enterocolitis (EC) is the most serious and potentially life-threatening complication of HD. It may present with a wide range of clinical presentations including abdominal distension, explosive diarrhea, vomiting, fever, lethargy, rectal bleeding, and shock.[21] The rates of postoperative enterocolitis vary from 0-66.66% in various published series of TEPT.[11,22,23][Table 3].

In few previous PTASPT studies, the rates of EC vary from 0-11.7%.[11,14,24-26] [Table 4]. In our series, post-operative EC occurred in 2 patients (8.3%). These patients recovered with aggressive approach with intravenous antibiotics and warm saline rectal wash-outs followed by regular anal dilatation. We had 2 (8.3%) mortalities in our series. One neonate who had a poor post-operative recovery succumbed

to overwhelming sepsis on the 5th post-operative day. The patient did not have any significant pre-operative problem or any eventful intra-operative period. The other patient expired after 3 months due to medical illnesses unrelated to the surgical problem or the procedure. Normal bowel function is the ultimate goal after surgery for HD. Out data is on fecal continence and bowel control is based on short-term follow-up. At 1 year follow-up and their respective for age follow-up, normal bowel habit was achieved in 18 patients (75%). Two patients who recovered from enterocolitis had ongoing minor problems such as loose stools and constipation requiring regular anal dilatation and occasional stool softeners at home. Another patient who had perianal fistula is on diversion colostomy awaiting further intervention. Urinary continence was noted in all the patients in our series who were old enough to be assessed and 80% of the parents who were able to be contacted confirmed that they witnessed spontaneous erections in the male patients post-operatively. Our data compared favorably with the other classical pull through procedures, TEPT,[17,27-31] and previous transanal Swenson's pull procedures.[11,14,20,24,25]

## CONCLUSION

Our short term data has shown that with appropriate skill and resources, primary transanal Swenson's procedure is a viable and safe option in all age groups including neonates in a developing country. It offers several social and financial advantages to the child and the family. We have also confirmed the feasibility of transanal pull-through for upper sigmoid colon HD as found in few previous reports. However, further studies documenting the long term results of this approach, particularly with respect to the incidences of gas bloating and enterocolitis and on urinary continence and sexual function will be needed as these children grow and develop.

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**How to cite this article:** Chawngthu V, Lalliandinga J. Primary Transanal Swenson's Pull-through in Hirschsprung's Disease in SRHF, Mizoram, India. Ann. Int. Med. Den. Res. 2019; 5(1):SG28-SG33.

**Source of Support:** Nil, **Conflict of Interest:** None declared