

LAPROSCOPIC ASSISTED SWENSON PULL-THROUGH FOR CLASSIC HIRSCHSPRUNG'S DISEASE WITH SIGMOID COLOSTOMY: A SINGLE CENTER EXPERIENCE

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ABSTRACT

OBJECTIVE: To share our experience of laparoscopic assisted Swenson pull-through for classic Hirschsprung's disease (HD) with sigmoid colostomy.

METHODS: Thirty-one patients with HD and sigmoid colostomy operated in Pediatric Surgery department, Khyber Teaching Hospital, Peshawar, Pakistan, between June 2014 and June 2018 were included. Patients with long segment HD, and those operated elsewhere were excluded from the study. Demographic characteristics, intraoperative findings and post operative course were retrieved from files. Post-operative outcome at 3rd, 6th and 12th month follow ups were taken from follow up charts.

RESULTS: Mean age and weight of patients were 13.23 ± 1.76 months and 7.89 ± 1.42 kg, respectively. Male to female ratio was 6.75:1. Mean operative time, duration of urinary catheterization and hospital stay were 171.87 ± 15.76 min, 0.79 ± 1.02 days and 5.91 ± 1.01 days, respectively. After surgery, stool consistency was liquid in 12 (61.29%) and loose in 12 (38.7%) patients. However, stool frequency was >3 times/day in 10 (32.25%) cases, 2-3 times/day in 18 (58.06%) patients and 0-1 time/day in 3 (9.67%) patients. Surgical site infection and perianal excoriation were seen in 2 (6.45%) and 11 (35.48%) patients, respectively. Progressive improvement in stool consistency and frequency was noticed at 12th month follow up. Perianal excoriation was seen in 11 (35.48%) patients at 3rd month, 6 (19.35%) patients at 6th and 1 (3.22%) patient at 12th month follow up.

CONCLUSION: Laparoscopic assisted Swenson pull-through is a good treatment option for patient with recto-sigmoid HD and bears a predictable and acceptable stooling pattern, optimum hospital stay and minimal complications.

KEY WORDS: Hirschsprung's Disease (MeSH); Colon, Sigmoid (MeSH); Colostomy (MeSH); Laparoscopic assisted pull-through surgery (Non-MeSH); Swenson pull-through (Non-MeSH).

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constipation is due to decreased gut motility secondary to congenital absence of ganglion cells in the involved portion of gut. The disease mostly presents in infancy but patient can report at any age usually before of 10 years.⁸ A barium enema can give a preliminary diagnosis of Hirschsprung's disease but deficiency/absence of ganglionic cells on rectal biopsy is a confirmatory.^{9,10} Our study included only those patients which had classic recto sigmoid HD.

Treatment of the condition is surgical and involve resection of proven aganglionic colonic segment and anastomosing the remaining. Soave, Boley, Duhamel and Swenson are different surgical procedures.¹¹ In Swenson pull-through, the aganglionic segment of colon is resected and ganglionic portion is pulled through and anastomosed 1.5 cm above the dentate line. Orvar Swenson devised this procedure in 1940s, so is named after him.^{12,13} With the advent of minimal invasive surgery, pull-through procedures are performed laparoscopically and even with robot. As there is no local study available on laparoscopic assisted pull-through surgery, we planned to share our experience of laparoscopic assisted Swenson pull-through in patients with classic Hirschsprung's disease involving the rectosigmoid gut only. Since we do not have the facility of frozen section biopsy, prior sigmoid colostomy is a pre-requisite for laparoscopy assisted Swenson pull-through in our set up.

METHODS

This retrospective study was conducted at

INTRODUCTION

The history of Hirschsprung's disease (HD) is traced back to Frederik Ruysch, a Dutch anatomist, who reported it in 1691.¹ However, the disease is named after a pathologist, Harald Hirschsprung, who first presented the histopathologic images of this disease in 1887 in two infants.² The literature reveals the incidence of HD as 1 in 5000 live births.^{3,4} It is seen four times more commonly in males as compared to females and is

predominant in white ethnicity.⁵ The condition has a significant association with Down syndrome with an incidence of 9%.⁶

Hirschsprung's disease is a hereditary condition of large gut and RET proto-oncogene is a predominant genetic culprit.⁷ It is congenital aganglionosis of colon in an ascending pattern involving rectum and in all cases and may be extensive in a proximal fashion up the large gut or less commonly small gut too. The hallmark presentation of chronic

TABLE I: OPERATIVE OUTCOME OF LAPAROSCOPIC ASSISTED PULL-THROUGH SURGERIES DURING HOSPITAL STAY (n=31)

Operative outcomes		No. of patients (%)
Stool consistency	Liquid	19 (61.29%)
	Loose	12 (38.70%)
	Formed	0 (0.0%)
Stool frequency	0 to 1 times/day	3 (9.67%)
	2 to 3 times/day	18 (58.06%)
	More than 3 times/day	10 (32.25%)
Surgical site infection		2 (6.45%)
Perianal excoriation		11 (35.48%)
Enterocolitis		0 (0.0%)
Adhesive obstruction		0 (0.0%)
Mortality		0 (0.0%)

TABLE II: FOLLOW UP DATA AT 3RD, 6TH AND 12TH MONTH OF LAPAROSCOPIC ASSISTED PULL-THROUGH SURGERIES (n=31)

Parameters		At 3 rd month	At 6 th month	At 12 th month
		n (%)	n (%)	n (%)
Stool consistency	Liquid	0(0.0%)	0(0.0%)	0(0.0%)
	Loose	18 (58.06%)	3 (9.67%)	0(0.0%)
	Formed	13 (41.93%)	28 (90.32%)	31 (100.0%)
Stool frequency	0 to 1	6 (19.35%)	15 (48.38%)	24 (77.41%)
	2 to 3	17 (54.83%)	10 (32.25%)	7 (22.58%)
	More than 3	8 (25.80%)	6 (19.35%)	0(0.0%)
Perineal excoriation		11 (35.48%)	6 (19.35%)	1 (3.22%)
Enterocolitis		0(0.0%)	0(0.0%)	0(0.0%)
Adhesive obstruction		0(0.0%)	2(6.45%)	0(0.0%)
Mortality		0(0.0%)	0(0.0%)	0(0.0%)

Pediatric Surgery department, Khyber Teaching Hospital, Peshawar, Pakistan. Study was approved by institutional review and ethical board. Thirty-one patients with confirmed Hirschsprung's disease (HD) on histopathology and sigmoid colostomy operated in between June 2014 and June 2018 were included in the study. HD patients with acute enterocolitis, massively dilated rectum (delayed referral) and long segment (transition zone above the sigmoid) were excluded from the study. All patients had primary decompressing colostomy before laparoscopic assisted pull-through procedure was offered.

After formal gut preparation and intravenous antibiotics at the induction of anesthesia, Indwelling catheterization and nasogastric intubation were done. Trendelenburg was our preferred position. Umbilical port of 3 mm was always our first port for pneumoperitoneum while 2nd and 3rd 3 mm ports were placed under laparoscopic guidance in right and left flank. Rectal dissection was done distally

till the peritoneal reflection and colonic mobilization upto splenic flexure proximally by Ligasure (Figure 1). Rest of the pull-through was done by transanal approach (Figure 2). Aganglionic portion was resected and ganglionic portion was anastomosed above the dentate line. The nasogastric tube was usually removed on the 2nd postoperative day and oral feeding started.

Demographic features, history and physical examination were noted. Intraoperative findings of patients undergone laparoscopic assisted pull-through procedure were recorded. The data was entered into SPSS version 20, computer program and analyzed accordingly. Study variables were analysed by simple descriptive statistics. Mean and standard deviation were calculated for numerical variables (age). Frequency and percentage were calculated for gender, intraoperative findings and post-operative outcome of laparoscopic assisted pull-through procedure at 3rd, 6th and 12th month follow ups

RESULTS

Out of 31 patients, 27 (87.09%) were males and 4 (12.90%) were females with male to female ratio of 6.75:1. Patients ranged in age from 9–18 months and mean age of patients was 13.23 ± 1.76 months. Mean weight of babies was 7.89 ± 1.42 kg. There were no case of any other associated congenital abnormality. There was no past history of surgery other than sigmoid colostomy in all patients.

All cases were successfully completed laparoscopically and none of the cases was converted to open surgery. Mean operative time of laparoscopic surgery was 171.87 ± 15.76 minutes. Mean duration of urinary catheterization was 0.79 ± 1.02 days. Mean hospital stay was 5.91 ± 1.01 days. Surgical site infection was noted in 2 (6.45%) cases. We did not encounter any mortality. The Operative outcome of laparoscopic assisted pull-through surgeries during hospital stay and Follow up at 3rd, 6th and 12th month of laparoscopic assisted pull-through surgeries are shown in Table I and II respectively.

DISCUSSION

Regarding our findings, the mean age of patients was 13.23 ± 1.76 months (Range: 9–18 months). In a study by Georgeson KE, et al. the age of patients ranged from 3 days to 966 days.¹⁴ In another study by Yan J, et al. the mean age of patients who underwent laparoscopic surgery for Hirschsprung disease was 6.2 ± 2.07 months.¹⁵ Similarly in work by Singh S, et al. the mean age of patients was 6.0 ± 0.23 months.¹⁶ Male dominance was observed in our study i.e. 87.09% male and 12.90% females. Males were also predominant in a study by Yan J, et al. i.e. 72.23 % male.¹⁵ As far concerned weight, the mean recorded weight was 7.89 ± 1.42 kg in our study. Similarly, in a study by Yan J, et al. the mean weight of patients undergoing laparoscopic surgery for Hirschsprung disease was 8.1 ± 2.45 kg.¹⁵ Study by Singh S, et al. found male to female ratio of 24:1.¹⁶ We did not come across any other congenital abnormality (0.0%) in association to the condition under consideration. However, in a study by Singh S, et al. 4.0% subjects exhibited features of Down syndrome.¹⁶

The mean recorded operative time of laparoscopic assisted pull-through was 171.87 ± 15.76 min in our study. In a study

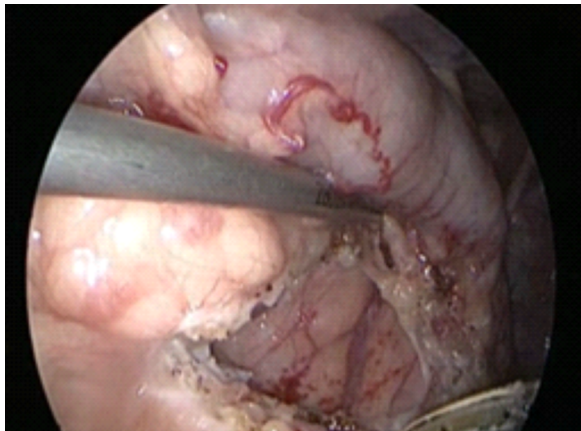


Figure 1: Laparoscopic mobilization



Figure 2: Transanal pull-through

by Georgeson KE, et al. the mean duration of surgery was 2.5 hours.¹⁴ Similarly, in a study by Yan J, et al. this figure was 175.5 ± 20.7 min.¹⁵ Similarly Singh S, et al. showed a mean operative time of 80 ± 0.30 min.¹⁶ Not a single laparoscopic pull thorough surgery was converted to open surgery (0.0%) in our study. However, in a study by Georgeson KE, et al. conversion to open surgery was reported in 2.5% patients.¹⁴ Similar to our study, there was no conversion of laparoscopic assisted surgery to open surgery in any patient in a study by Singh S, et al.¹⁶

We recorded a mean duration of urinary catheterization as 0.79 ± 1.02 days. Post operatively during hospital stay, the consistency of stool was liquid 19 (61.29%) patients while in other 12 (38.70%) patients the consistency was stool was found loose in our study. However, formed stool consistency was not observed in any patient i.e. 0.0%. In a study by Georgeson KE, et al. 7.5% patients developed diarrhea after surgery.¹⁴ We graded stooling frequency as time per day and post operatively we recorded a frequency of 0-1 times/day in 3 (19.35%) cases, 2-3 times/day in 18 (54.83%) and more than 3 times/day in 10 (32.25%) patients. Surgical site infection was recorded in 2 (6.45%) patients in our study. However, surgical site infection was not seen in any patient (0.0%) after laparoscopic surgery for Hirschsprung disease in a work by Yan J, et al.¹⁵ In our study, 11 (35.48%) patients developed perianal skin excoriation in post operative hospital stay. Mean hospital stay of patients in our study was 5.91 ± 1.01 days. In a study by Georgeson KE, et al. the average hospital stay was 3.7 days.¹⁴ Similar to our

study, the mean operative time of laparoscopic surgery for Hirschsprung disease was 6.2 ± 1.5 days in a study by Yan J, et al.¹⁵ Another study by Singh S, et al. showed this duration as 6 ± 0.21 days.¹⁶

The follow up data of patients at 3, 6 and 12 months was retrieved (Table II). Consistency of stool at 3rd month follow up was loose in 18 (58.06%) patients. However, most of these patients showed improved consistency towards formed stools at 6th and 12th month follow up i.e. 18 (90.32%) and 31 (100.0%) patients, respectively in our study. The stool frequency was high in majority of patients at 3rd month follow up but at 6th and 12th month follow ups, was improved (decreased) in most of these patients at 6th and 12th month. Perianal skin excoriation was seen in 11 (35.48%) patients at 3rd month follow up, 6 (19.35%) patients at 6th month follow up and 1 (3.22%) patients at 12th month follow up in our study. In a study by Yan J, et al. the diaper rash was seen in 5.56% patients post laparoscopic surgery for Hirschsprung disease at 6-12 months follow up.¹⁵ Enterocolitis was not observed in any patient at all follow ups in our study, however, adhesive intestinal obstruction was diagnosed in 2 (6.45%) patients at 6th month follow up which was managed conservatively. In a study by Georgeson KE, et al. enterocolitis was reported in 7.5% patients.¹⁴ However, enterocolitis and intestinal obstruction were not observed in any patient after laparoscopic surgery for Hirschsprung disease in a study by Yan J, et al.¹⁵

We did not encounter any mortality (0.0%) throughout our study. Similarly, in a study by Georgeson KE, et al. no

mortality was observed.¹⁴ However, mortality (4.0%) was observed in a study by Singh S, et al.¹⁶

LIMITATIONS

This study has certain limitations as it is a single center study with limited patients. A large series of patients is required to thoroughly evaluate the results of the modality discussed.

CONCLUSION

Heterogeneity in results of different modalities of treatment like Swenson, Soave and Duhamel pull-through procedures is preventing identification of a gold standard. Laparoscopic assisted Swenson pull-through is a recent advancement in minimal invasive approach for the treatment of HD. Our experience with this technique shows that it can be a good treatment option for patients with HD. However the procedure needs to be compared with its open alternative and other procedures for functional outcome and complications prior to recommending it.

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AUTHORS' CONTRIBUTIONS

Following authors have made substantial contributions to the manuscript as under:

SA: Conception & study design, acquisition of data, drafting the manuscript, critical review, approval of final version to be published

IUR: Acquisition of data, drafting the manuscript, approval of final version to be published

TW & MI: Analysis and interpretation of data, drafting the manuscript, approval of final version to be published

FA: Acquisition, analysis and interpretation of data, critical review, approval of final version to be published

HA: Study design, acquisition, analysis and interpretation of data, drafting the manuscript, approval of final version to be published

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

CONFLICT OF INTEREST

Authors declared no conflict of interest

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NIL

DATA SHARING STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.



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