

## Outcome of single-stage transanal Swenson pull-through without frozen section in short-segment Hirschsprung's disease

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### Abstract

**Objective:** To evaluate the outcome of single-stage transanal Swenson pull-through without frozen section in paediatric patients with short-segment Hirschsprung's disease.

**Method:** The single-group, prospective, cohort study was conducted at the Paediatric Surgical Unit, Nishtar Medical University, Multan, Pakistan, from January 2020 to December 2022 and comprised patients aged 1-12 years who were confirmed to have Hirschsprung's disease through rectal biopsy and demonstrated short-segment involvement on contrast enema. Single-stage transanal Swenson's pull-through procedure was performed on all patients without the use of frozen sections. Postoperative follow-up lasted 6 months. Data was analysed using SPSS 25.

**Results:** Of the 97 patients, 64(66%) were boys and 33(34%) were girls. The overall mean age was  $2.97 \pm 1.62$  years. Complications were observed in 21(21.65%) patients, with anastomotic stricture being the most common 6(6.19%), followed by increased stool frequency 5(5.15%), enterocolitis 5(5.15%), and anastomotic leakage 3(3.09%). Biopsy of the excised segment revealed that 2(2.06%) patients had aganglionic bowel pull-through.

**Conclusion:** The single-stage transanal pull-through, when performed in the absence of frozen section, was found to be safe in the management of short-segment Hirschsprung's disease.

**Key Words:** Hirschsprung disease, Swenson pull-through, Transanal pull-through, Anastomotic stenosis, Enterocolitis.

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### Introduction

The absence of ganglion cells in the intrinsic nervous system of gastrointestinal (GI) tract is salient feature of Hirschsprung's disease (HD). It mostly affects the distal part of the colon, but the length of the affected bowel segment remains variable. About 80% of the children have short-segment disease where only the recto-sigmoid colon is deficient of ganglion cells.<sup>1</sup> HD has variable presentation, and it is a key differential for chronic constipation in paediatric population.<sup>2</sup>

Definitive surgical management includes resection of dysfunctional bowel and re-establishing intestinal continuity, which is called the Swenson pull-through operation. Various single-stage (primary) or multi-stage procedures, performed by open or laparoscopic technique, are in clinical practice.<sup>3</sup> Recently, the single-stage transanal pull-through has gained worldwide popularity. The single stage, avoidance of stoma with

relevant complications, decreased hospital admissions, and reduced post-surgical complications are factors in favour of the single-stage transanal pull-through<sup>4</sup>. Variable outcomes of the single-stage transanal pull-through are mentioned in literature.<sup>4,5</sup> Most studies have supported this approach, but some authors have suggested that the transanal approach may damage the sphincter complex, leading to poor functional outcome.<sup>6</sup>

Intra-operative frozen section is an essential part of the single-stage transanal pull-through procedure. It is used to define the extent of surgical resection. It increases the operative time and the cost of the procedure. It also requires skilled personnel. In developing countries, like Pakistan, due to limited resources and the non-availability of frozen section facilities and skilled personnel, many people have challenged the use of intraoperative frozen section.<sup>7</sup> Different centres have reported their results for the procedure with varying success and complication rates.<sup>7,8</sup>

The current study was planned to measure the outcome of the single-stage transanal Swenson pull-through procedure without frozen section in paediatric patients with short-segment HD.

### Patients and Methods

The single-group, prospective, cohort study was

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conducted at the Paediatric Surgical Unit, Nishtar Medical University, Multan, Pakistan, from January 2020 to December 2022. After approval from the institutional ethics review board, the sample size was calculated using the standard formula for proportions in single-group studies:  $n = (Z^2 \times P \times (1-P)) / d^2$ , where  $Z = 1.96$  (corresponding to 95% confidence level),  $P = 0.10$  (anticipated complication rate based on previous literature<sup>9</sup>, and  $d = 0.06$  (absolute precision/margin of error). The sample was raised using convenience sampling technique. Informed consent was taken from parents of all the enrolled subjects.

Those included were patients aged 1-12 years who were confirmed to have HD through rectal biopsy and demonstrated short-segment involvement on contrast enema. Those with associated comorbidities, congenital syndromes, a history of prior surgical intervention, stoma formation, non-responsiveness to rectal washouts and whose parents were non-compliant with rectal washes or unwilling to consent to the procedure were excluded.

Patient demographics were recorded. The attendants were trained for rectal irrigation with normal saline (20ml/kg) at home once daily till the time of surgery. The duration of rectal irrigation depended on colonic dilatation. The duration of rectal irrigation was extended in patients with a more dilated proximal colon to decrease colonic diameter. All patients were admitted two days before surgery and placed on a clear liquid diet. Mechanical bowel preparation was achieved by saline irrigations. A triple antibiotic combination (Amoxicillin 100mg/kg, Cefotaxime 50mg/kg and Metronidazole 7.5mg/kg) was administered intravenously one day before surgery and repeated three times a day during the postoperative course for 3-5 days depending upon the patient's clinical status and tolerance of oral feed.

A single-stage transanal Swenson pull-through without frozen section was performed in all patients. Resection of the aganglionic segment with narrow lumen on gross appearance was continued. After identifying the transition zone, dissection was continued at least 5cm proximally on dilated and hypertrophied intestine, which was considered normal ganglionic bowel. Intestinal continuity was re-established with single layer anastomosis about 0.5cm above the dentate line. The excised specimens were submitted for histopathological evaluation.

Oral feeding of transparent non-stool-forming liquids was started after 72 hours, with advancement to a routine diet the next day. The patients were discharged after establishing oral feed. Follow-up visits were scheduled at

2 weeks, 6 weeks, 3 months and 6 months. However, postoperative outcomes were assessed and reported based on findings at the final 6-month follow-up.

Postoperative anal dilatation was not started unless required for anastomotic stricture. The patients were regularly monitored during admission and at each follow-up visit. Data about patient characteristics and outcome variables, including operative time, anastomotic leakage, length of hospital stay (LOS), return to feeding, frequent stooling (>3 times per day), persistence of symptoms (constipation, abdominal distension), anastomotic stricture, and histopathology results, was maintained on a pre-designed proforma.

Data was analysed using SPSS 25. Mean  $\pm$  standard deviation were calculated for continuous data, while frequencies and percentages were calculated for categorical data. Chi-square test was used to compare post-stratification values with respect to age and gender.  $P < 0.05$  was considered statistically significant at 95% confidence interval. In instances where the expected cell frequency was  $< 5$  in  $> 20\%$  cells, Fisher's exact test was applied. Independent sample t-test was used to compare continuous variables between patients with and without complications.  $P < 0.05$  was considered statistically significant.

## Results

Of the 97 patients, 64(66%) were boys and 33(34%) were girls. The overall mean age was  $2.97 \pm 1.62$  years (range: 0.64-6.76 years). The mean weight was  $12.99 \pm 5.08$  kg (range: 7-33kg). The mean haemoglobin (Hb) was  $10.92 \pm 1.48$  g/dl (range: 8.5-15 g/dl). The mean operative time was  $119.6 \pm 11.76$  minutes (range: 90-150 minutes). The mean LOS was  $7.18 \pm 1.0$  days (range: 6-12 days).

Complications were observed in 21(21.65%) patients, with anastomotic stricture being the most common 6(6.19%), followed by increased stool frequency 5(5.15%), enterocolitis 5(5.15%), and anastomotic leakage 3(3.09%).

**Table-1:** Complications at 6-month follow-up (n=97).

Complication	Responses	
	No. of Patients	Percentage %
Anastomotic leakage	3	3.09
Anastomotic Stricture	6	6.19
>3 Stools per day	5	5.15
HAEC	5	5.15
Persistent Obstructive Symptoms	0	0
Aganglionic Pull through	2	2.06
Death	0	0
Total	21	21.65

HAEC: Hirschsprung-associated enterocolitis.

**Table-2:** Complications stratified by gender.

Complication	Male(Total=64)		Female(Total=33)		P-Value
	No. of Patients	Percentage %	No. of Patients	Percentage %	
Anastomotic leakage	2	3.12 %	1	3.03 %	.980
Anastomotic Stricture	2	3.12 %	4	12.12 %	.081
>3 Stools per day	3	4.68 %	2	6.06 %	.772
HAEC	3	4.68 %	2	6.06 %	.772
Aganglionic Pull through	1	1.56 %	1	3.03 %	.630
Total	11	17.18 %	10	30.30 %	

HAEC: Hirschsprung-associated enterocolitis.

**Table-3:** Complications stratified by age groups.

Complication	Age Groups				P-Value
	1 Year to 5 Years ( Total=90)		>5 Years to 12Years (Total=7)		
	No. of Patients	Percentage %	No. of Patients	Percentage %	
Anastomotic leakage	3	3.33%	0	0	.203
Anastomotic Stricture	6	6.67%	0	0	1.00
>3 Stools per day	4	4.44%	1	14.29%	1.00
HAEC	5	5.56%	0	0	1.00
Aganglionic Pull through	2	2.22%	0	0	1.00
Total	20	22.22%	1	14.29%	

Hirschsprung-associated enterocolitis.

Fisher's exact test was used where Chi-square test assumptions were not met (expected cell counts <5 in more than 20% of cells)

Biopsy of the excised segment revealed that 2(2.06%) patients had aganglionic bowel pull-through. No deaths were reported (Table 1). No significant association of outcome variables with gender (Table 2) and age groups (Table 3) was identified.

No statistically significant differences were observed between patients with and without postoperative complications in terms of age ( $p=0.952$ ), weight ( $p=0.118$ ), Hb level ( $p=0.522$ ), operative time ( $p=0.231$ ), or LOS ( $p=0.564$ ).

Patients who developed anastomotic leakage were managed by ileostomy in early postoperative period. Patients who developed anastomotic stricture responded to anal dilatation without surgical intervention. Patients with aganglionic segment pull-through were managed with staged procedure later on.

## Discussion

The surgical management of HD has progressively evolved over the years, transitioning from being managed as an open, multi-staged procedure to a minimally invasive, single-stage procedure.<sup>4,10</sup> Recently, the trend has shifted towards transanal pull-through procedures to avoid adverse events associated with abdominal incisions. It also reduces operative time and the total length of hospital stay. In the study, the mean

operative time was 119.6 minutes, and the mean LOS was 7.18 days. These results are comparable to earlier findings.<sup>7,11,12</sup>

Biopsy is an essential part of pull-through procedures because it helps determine the level of aganglionosis, and defines the extent of surgical dissection. It is performed by open or laparoscopic techniques before the pull-through procedure or intra-operatively using frozen section. Single-stage transanal pull-through procedures are ideally performed with intraoperative frozen sections. In developing countries, like Pakistan, this approach is challenged due to limited resources and the non-availability of frozen sections and skilled personnel. Moreover, differences between intraoperative frozen sections and final histopathology results have also been reported.<sup>13</sup>

In the current study, dissection was initiated approximately 1cm above the dentate line, and, after identifying the transition zone, dissection continued at least 5cm proximally on the dilated bowel wall. Histopathology results of 95(97.9%) patients were normal in the current study, while 2(2.06%) patients had aganglionic or transition zone pull-through. A recent study in Peshawar reported ganglionic bowel pull-through in all patients with a minimum 5cm dissection proximal to the transition zone.<sup>8</sup>

In the literature, variable success and complication rates have been reported.<sup>8,14,15</sup> In the current study, 21(21.65%) patients developed complications, which is almost the same as mentioned in other studies. In the literature, a higher complication rate is associated with small age and male gender.<sup>8</sup> In this study, gender distribution was the same as described in the literature for HD<sup>15,16</sup>, but no significant association of complications with age and gender ( $p > 0.05$ ) was identified.

In the early postoperative period, anastomotic leak is a serious complication after a pull-through procedure. In the current study, 3(3.09%) patients had an anastomotic leak. In these patients, an ileostomy was created as an emergency procedure, and redo surgery was performed thereafter. A 2.6% and 3% incidence of anastomotic leak is reported in the literature.<sup>14,17</sup>

Anastomotic stricture was the most common complication reported in 6(6.19%) of the current patients. The reported incidence of anastomotic stricture in other studies is 9%.<sup>15</sup> Ischemia or undue tension at anastomotic line is considered to be the underlying factors for stricture development.<sup>18</sup> All such patients were managed with anal dilatation without the need for another surgical procedure.

Enterocolitis is a major life-threatening complication in patients with HD, and can occur both before and after the definitive surgical repair.<sup>19</sup> In the current study, 5(5.15%) patients suffered enterocolitis. The incidence of enterocolitis was in a 2022 study reported to be 10%, and it was 14.3% in another study.<sup>20,21</sup>

The current study has its limitations of a small.

sample size and selection bias due to consecutive sampling. Besides, it was a single-centre study which did not assess long-term outcomes. Multicentre randomised control trials (RCTs) are needed to validate the current findings.

## Conclusion

The single-stage transanal Swenson pull-through, when performed in the absence of frozen section, was found to be safe in for the management of short-segment HD with minimal complications and a comparable success rate. Resection >5cm proximal to the transition zone ensured a normal ganglionic bowel pull-through.

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**AUTHOR'S CONTRIBUTION:**

**KTK:** Conceived, collected data, wrote synopsis, reviewed, final approval and agreement to be accountable for all aspects of the work.

**ARR:** Conceived, preparation, final approval and agreement to be accountable for all aspects of the work.

**MAH:** Design, critical review, final approval and agreement to be accountable for all aspects of the work.