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ORIGINAL ARTICLE

Single Stage Transanal Pull through in patients with Short Segment Hirschprung's Disease-Our Experience with Swenson Technique

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ABSTRACT

Background: To study the results of primary transanal pull-through in the patients of short-segment Hirschsprung's disease (HD) without using the frozen section facility.

Patients and methods: This retrospective cohort study was performed in the Department of Paediatric Surgery, Nishtar Medical University, Multan, from January 2021 to December 2023. Thirty-five patients of more than one year of age, diagnosed with short segment HD based on history, examination, barium enema, and rectal biopsy, were included in the study. Patients having HD with any syndrome or with stoma were excluded from the study. One-stage transanal Swenson pull-through was performed, and the transition zone was identified on naked-eye visualization. No frozen section facility was used. Patients were followed for six months for complications and outcomes.

Results: Out of 35 patients, 8 (22.8%) had a total of 11 complications. Enterocolitis was most frequent, seen in 3 (8.6%) patients. Other complications included anastomotic stricture, anastomotic leakage, constipation, fecal incontinence, and abdominal distension. Two biopsies showed an absence of ganglion cells, and it was significantly associated with complications (p-value .007). Two patients needed diversion stoma. Overall, 27 (77.14%) patients showed good results post-operatively.

Conclusion: Primary transanal endorectal pull-through can be performed safely in children with short-segment HD without the use of frozen section facilities. Careful identification of the transition zone during the surgery is important, as inadequate resection can result in multiple complications. However, most of the complications can be managed conservatively.

Keywords: Hirschsprung's disease, Transanal pull through, Frozen section, Enterocolitis.

INTRODUCTION

Hirschsprung's Disease (HD) is an embryological disease of the intrinsic part of the enteric nervous system characterized by absent ganglion cells, affecting 1 in 5000 live births. 80% of the children with HD have short-segment HD where only the rectosigmoid colon is deficient of ganglion cells[1]. Neonates present with clinical signs of distal intestinal obstruction shortly after birth. However, a history of delayed passage of their 1st meconium and severe constipation is the predominant symptom in older children. Diagnosis is through radiography, manometry, or different type of biopsies[2].

Surgical treatment of HD includes removal of the aganglionic bowel segment and re-establishing intestinal continuity through anastomosis of the ganglionic bowel to the anus. Various single- or multi-staged pull-through procedures can be used for this purpose. The endorectal pull-through performed either transanally (TERPT) or in combination with laparotomy or laparoscopy (LERPT) has gained popularity recently. Due to the lack of any abdominal incision or transabdominal dissection, transanal endorectal pull-through is considered the least invasive. It obviously leaves no scars[3]. After the introduction of TERPT in 1998, several studies have shown its favorable results along with fewer complication rates[4]. The benefits of TERPT include no need for colostomy, easy technique, low bleeding rate and short hospital stay compared to other techniques[5]. Potential risks include fecal incontinence, colonic torsion, and the inability to identify the histological transition zone[6].

According to literature, the main prerequisite for a single-stage transanal pull-through is the availability of a frozen section facility to determine the level of aganglionosis during surgery. It may be freely available in western countries, but in low- and middle-income countries (LMICs), machines as well as trained personnel may not be available everywhere. Therefore, TERPT is sometimes deferred as an option in the definitive management of short segment HD. However, some authors have reported doing TERPT without the use of frozen section. Such studies are sparse, yet results are encouraging, though not without complications. Choudhury et al. from Bangladesh showed that thirty-nine children out of 41 appeared to have excellent results and did not have any complications[7]. Another study showed that only 20.4% of patients showed some sort of complications[8].

The TERPT was performed in all these studies without using frozen section facility. Like many other hospitals in LMICs, our hospital lacks the facility of frozen section. Despite that, we perform TERPT quite often. There is a lack of local data, especially data from our region, about the results of primary transanal pull-through without the use of a frozen section facility, so this study was planned to fill this research gap and to share results of primary TERPT without the use of a frozen section in patients of short segment HD.

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METHODOLOGY

This retrospective cohort study was conducted in the department of Pediatric Surgery, Nishtar Medical University, Multan. After approval from the ethical review board (IERB no. 7079, dated 03-06-2024), patients' records were collected from January 2021 to December 2023. Patients with a short segment HD were selected. Patients from both genders, aged between one year and twelve years, were included in the study. Diagnosis was made based on barium enema in an unprepared gut and rectal biopsy. Patients with stoma, long segment disease, HD with any syndrome, a history of previous surgery, and HD with any other medical problem were excluded from the study. 35 patients were included in the study.

Surgical procedure

The patients less than one year of age at presentation or with gross dilatation of the colon were kept on daily saline enemas (20 ml per kg) at home till age was one year or colonic dilatation was resolved. All patients were admitted three days before surgery for final preparation. Mechanical bowel preparation was achieved with saline irrigations. The diet was restricted 72 hours before surgery, and clear fluids were started 12 hours before surgery. Triple antibiotic coverage (gram positive, gram negative, and anaerobes) was started 24 hours before surgery and continued till the 5th postoperative day. After on-table lavage, transanal Swenson pull-through was performed in a supine position. Based on the picture of the barium enema, the aganglionic segment was decided on gross appearance, having thickened muscles and a narrow lumen.

The dissection was carried proximally till the transition zone, identified by the dilated part of the colon. Further, 2 to 3 centimeters were also mobilized before resection, and the proximal part was brought down to be anastomosed with the anus about one centimeter above the dentate line. The resected segment was submitted for histopathology. Clear fluids started after 24-48 hours. Solid foods gradually started after the 3rd postoperative day. Patients were discharged after 5-7 days if the postoperative period was uneventful. Post-operative dilatation was not started until a stricture was suspected. Follow-up was scheduled for four weeks weekly, fortnightly for next month, and monthly for the next four months.

Statistical analysis

Along with demographic data, the duration of surgery, spontaneous passage of stool, abdominal distension, bleeding per rectum, perineal skin excoriation, soiling, mucosal prolapse, constipation, anastomotic stricture (need for dilatation), biopsy report, anastomotic leak, enterocolitis, hospital stay, return to full oral feeding, fecal incontinence, and persistence of symptoms were recorded. All data was recorded on a proforma and analyzed using SPSS version 20. Quantitative data like age, weight, and duration of surgery were reported as mean and standard deviation. Categorical data, like complications, were expressed in frequencies and percentages. The Chi-square test was used to explore the association between different qualitative variables. The P-value of .05 was taken as significant. Tables are used to present the data.

RESULTS

35 patients were included in the study, including 24 (68.6%) male and 11 (31.4%) female. The mean age of the patients was 3.58 years ± 2.19 years. The minimum age of the patients was one year, and the maximum age was 8.25 years. The average weight was 14.54 ± 4.56 kilograms. The mean operative time was 140 minutes ± 17.03 minutes. Based on the clinical picture, clear fluids were started on the 2nd day, and gradually solid food was introduced. Return to full feed was on average after 4.80 days ± 1.32 days.ar relationship was found between age and length of surgery (Figure 2). The mean length of the stay (LOS) at the hospital was 6.60 (± 1.21) days, with a minimum stay of 5 days and a maximum stay of about 10 days (Figure 1). A linear relationship was found between age and length of surgery (Figure 2).

In the post-operative period, 91.4% (32) patients passed stool spontaneously, whereas 8.6% (03) patients passed stool with assistance (rectal stimulation by insertion of rectal drain). Histopathology examination of the proximal part of the excised bowel segment revealed the presence of ganglion cells at the anastomotic margin in 94% (33) of the patients, and ganglion cells were not identified in 6% (2) of the patients. Absence of ganglion cells and presence of hypertrophic nerve fibers were noted in the distal part of all excised specimens.

Among the different complications (Table 1), enter colitis was encountered in 03 (8.6%) patients. Anastomotic stricture was seen in 02 (6%) patients. 01 (3%) patient suffered from anastomotic leakage. It was initially dealt with rectal drain but later had to be managed with diversion ileostomy. Another patient whose report came back negative for ganglion cells also needed diversion stoma. Effort was made to identify association seen complications, sex, age the presence of ganglion cells on histopathology. Complications were not significantly associated with age other age patients (p-value 0.619) or sex of patient (p-value 0.656). However, a significant association was seen between the absence of ganglion cells and complications (p-value .007) (Table 2). Overall complications were seen in 8 (22.8%) patients (table no. 2). It is to be noted that the total number of complications was 11(3111 (31.4ut those were seen in total patients. 03 patients had more than one complication. That means 27 (77.14%) patients did well during the early follow-up period.

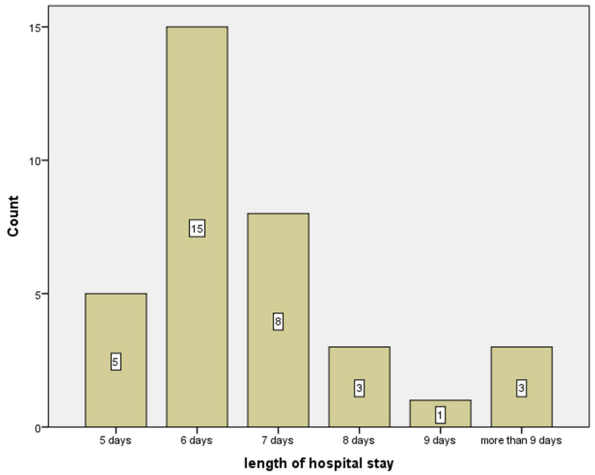

Figure 1. Length of Hospital stay (days)

Table 1: Post operative Complications

Complication	Number	Percentage
Abdominal distension	1	2.9
constipation	2	5.7
Fecal incontinence	2	5.7
Anastomotic leakage	1	2.9
Anastomotic stricture	2	5.7
Enterocolitis	3	8.6
Total	11	31.4

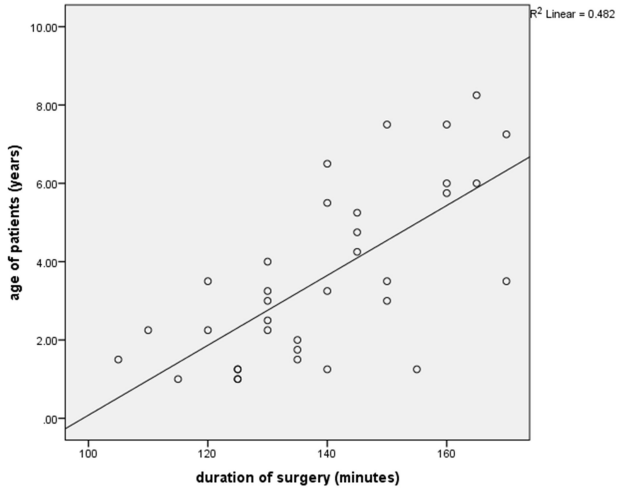

Figure 2. Association between age of patients & duration of Surgery (minutes)

Table 2: Gender & Histopathology association with complications

		complication		p-value
		yes	no	
gender of patient	male	6	18	.656
	female	2	9	
		complication		p-value
		yes	no	
ganglion cells	present	6	27	.007
	absent	2	0	

DISCUSSION

Single-stage transanal pull-through has become the procedure of choice for definitive treatment of Hirschsprung's disease, especially in the short-segment variety of the disease[9]. The main aim of our study was to share results of primary transanal pull-through without the use of frozen section and look for its feasibility

in resource-constrained settings. The identification of the length of the diseased portion was based on preoperative contrast study and naked-eye visualization during the surgery. In the absence of frozen section, the transition zone can be identified by contrast studies, 2D or 3D imaging, and/or laparoscopy[10-12], and single-stage transanal pull-through can be carried out safely[13]. Farooq et al. concluded that when the transition zone is reached, the surgeon must go 5 cm higher to ensure that the resection is not made in the transition zone[8]. We did not use a 5 cm cut-off but dissected roughly 2-3 cm proximally from the transition zone. The proximal end of the resected gut was sent for histopathology, and it showed that 33 (94%) of specimens had ganglion cells; however, specimens of 02 (6%) patients showed absent ganglion cells. It was the factor that has a strong association with the complications (p-value .007). More proximal resection, as suggested by some authors, can be a solution to this problem.

Enterocolitis was the most common complication, and it was seen in 03 (8.6%) patients. Two responded to conservative management, while in one diversion ileostomy was needed. Different studies have reported enterocolitis in 10-15% of patients[6-7]. A study showed that irrespective of age, the level of hemoglobin may be a strong risk factor for the development of HAEC[14]; however, we didn't find any such relationship. Anastomotic stricture was encountered in 2 (5.7%). We didn't perform anal dilatation routinely, but it was kept for the patients who developed signs of stricture in the form of difficulty in passing stools and abdominal distension. It has been reported by multiple authors with rates varying from 3.9%[17] to 35%[15]. Strictures were managed with anal dilatations. Anastomotic leakage was seen in 01 (2.9%) patients. However, our patient responded to the trans-anastomotic nelton drain, though a study had shown that a stoma may be needed for management of leakage[16]. Constipation was seen in 2 (5.7%) patients. Others have reported constipation in 8%[17] and 25%[6] of their series. In our series, it was relieved by bowel management except in the one patient, whose pathology report was negative for ganglion cells. Constipation can be treated with re-operations or suitable nutritional regimens, though it can be seen years after the operation[18-19]. Fecal incontinence was seen in 2 (5.7%) of patients. It presented as soiling. Its reported incidence varies between zero percent and 55%[18-20]. In some studies, fecal incontinence was the most frequent problem[1]. According to a study, stretching during surgery may have caused it, and that's why in younger patients its incidence is found in greater numbers[15].

Mean operative time was 139 ± 17 minutes. The patients returned to full oral feed on average after 4.80±1.32 days. This correlates with most of the data[6-10]. Mean hospital stay was 6.6±1.22 days. Different authors have reported average hospital stays of 3-6 days[8,15]. It seems that no abdominal exploration and initiating early oral feed has really helped in reducing the length of hospital stay[9,21]. In our study there were 69% male and 31% female patients. Although the ratio of male to female patients was comparable to data, there are reports where an association was shown between post-operative complications and the gender of the patients[8,15]. We couldn't find such an association (p-value .656). The average age of the patients was 3.58 years ± 2.19 years. There was no statistically significant association between age of patient and complications (p-value .619). A study observed that poorer outcomes were seen in the patients who were operated on during the neonatal period[15]; however, the study differed from this observation in their research work[17]. A study from Iran has concluded that age between 3 months and 12 months can be a good time to perform definitive surgical treatment of HD[5]. A study from Korea found that if TERPT is performed in infancy after toilet training, the uncomfortable symptoms keep on improving with time[4].

Overall complications were 11 (31.4%), but those were seen in 08 (22.8%) of the patients. Overall complication rate varies from 7.1% to 53%[7,10]. Some studies have reported a complication rate around the figure of our study[8,15]. The complication rate seems high, but if it is seen thoroughly, it can be concluded that most of the complications were of short term. In our study, only two (6%) of patients' biopsies showed no ganglion cells in the proximal part of the resected part of the gut; those were the patients who suffered most of the complications and problems: abdominal distension, constipation, late return to oral feed, increased length of stay, etc. Certain studies have tried to compare different strategies to come up with reasons for complications. Laparoscopic versus complete transanal approach found no difference in outcomes[1,11]. Similarly, resection of 5 cm or more than 5 cm from the suspected transition zone may have resulted in a lower complication rate, but such maneuvers have not been able to make the results complication free[7,8].

In our study, the 25 patients (72%) did well postoperatively. 28% had some sort of complication. However, 33 (94.3%) had passed stool normally during the follow-up. It's a good outcome sign. Voluntary bowel movements were achieved by 82% of the patients in a study by[17]. Mostly good results are reported in patients undergoing TERPT without frozen section facility, and our study also demonstrated that short segment HD can be safely managed with TERPT even in the absence of frozen section facility.

The limitations of the study may be that it was a retrospective study with a small sample size and lacking long-term follow-up. However, encouraging results should prompt us and others for prospective RCT studies with larger sample sizes and the involvement of multiple centers and long-term follow-up to ascertain the feasibility of TERPT in the absence of a frozen section facility.

CONCLUSION

Primary transanal endorectal pull-through can be performed safely in children with short-segment HD without the use of frozen section facilities. Careful identification of the transition zone during the surgery is important, as inadequate resection can result in multiple complications. However, most of the complications can be managed conservatively.

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