Comparison of two techniques for single-stage treatment of Hirschsprung disease in neonates

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Citation

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Abstract

Introduction:
The aim of this study is to determine the incidence, morbidity and functional outcome of two techniques for Hirschsprung disease (single-stage Duhamel pull-through and tans-anal pull-through) in the neonatal age group.
Method:
All the neonates with Hirschsprung disease with short aganglionic segment (transition zone at recto-sigmoid junction) admitted during the last 6 years were included. A total of 44 cases were included in the study of which 24 were treated by Duhamel pull-through (group A) and 20 were treated with trans-anal pull-through (TEPT, group B). The time of operation, acceptance of first oral feed, duration of hospital stay, complications and anal continence were evaluated for both procedures.
Result:
There was no difference in age and sex and length of the aganglionic segment of bowel. The average operative time was 140-180 min. in group A and 120-170 min. in group B. Per-oral feed was started on the 5-7th day in group A and on the 5-6th day in group B. Hospital stay was longer in group A whereas complications were more common in group B and were found to be statistically significant. Anal continence was almost similar in both groups.
Conclusion:
Single-stage Duhamel pull-through operation is better than trans-anal pull-through operation in long-term follow-up although duration of hospital stay was shorter in TEPT.

INTRODUCTION

Hirschsprung disease affects one in 5,000 newborns. The diagnosis often is suspected when a newborn fails to pass meconium in the first 48 hours of life, has abdominal distension, or has vomiting. In older children, a history of chronic constipation may be the presenting complaint. The diagnosis is suspected by a barium enema suggesting a transition point and confirmed by rectal biopsy showing the absence of ganglion cells Surgery for Hirschsprung disease has changed significantly during the last 2 decades. Multistage surgery has progressed to open or laparoscopically assisted 1-stage repair. One-stage totally transanal procedure is the latest evolution in the management of Hirschsprung disease [1,2,3] . One-stage surgery for Hirschsprung disease is well established, and the results are comparable or better than after 2- or 3-stage operations [4,5] . The rationale for primary surgery in the neonatal period has been the potential benefit of avoiding a colostomy during the first months of life and establishment of colonic continuity early in the life. It has been speculated that this may enhance the development of normal continence. Neonatal 1-stage procedures also have been suggested to be associated with less cost and demand of resources. De la Torre-Mondragon and Ortega-Salgado [1] first described totally transanal endorectal pull-through (TEPT) in the newborn in 1998. This prospective study was undertaken to investigate the safety and efficacy of 1-stage TEPT technique as a definitive treatment for patients with HD in the neonatal age group and to compare it with single-stage Duhamel pull-through (DP) operation.

METHOD

All the cases of short-length Hirschsprung disease presented in the neonatal age group in the last 6 years were included in our study. Group A includes the patients of short-length (transitional zone at recto-sigmoid or proximal sigmoid region) Hirschsprung disease presented in the neonatal age

group during March 1999 – Feb. 2002 and treated by single-stage Duhamel pull-trough without protecting colostomy. The patients of short-length Hirschsprung disease presented in the neonatal age group during March 2002 – Feb. 2005 and treated with by single-stage transanal rectal pull-through were grouped in group B. A total of 44 patients were included in our series of which 24 in group A and 20 in group B (table 1).

Figure 1

Table 1: Showing comparative parameters in the two groups

	Group A (DP)	Group B (TEPT)
No. of cases	24	20
Age group	12 – 30 days	14 – 30 days
Sex	M (15), F (9)	M (13), F (7)
Length of aganglionic segment	Transitional zone at RS – 18 Transitional zone in PS – 6	Transitional zone at RS – 15 Transitional zone in PS – 5

The mainstay for diagnosis of Hirschsprung disease was history, clinical examination and radiological investigation (barium enema) preoperatively and frozen section examination per-operatively. All the patients were operated by a single surgeon. The patients were evaluated on the basis of per-operative time, commencement of oral feed, duration of hospital stay, complications, anal continence and follow-up records. Statistical analysis was done by using Fisher's exact test. An analogue scoring system [12] was used to provide a functional outcome score for each child, using a combination of questionnaire follow-up, patient interview, and case-note review.

Normal bowel habit – 1
Soiling < 1 per week – 2
Soiling > 1 per week – 3
Daily soiling or need of enema – 4
Major revision surgery – 5

Satisfactory outcome was defined as a score of 1 or 2. Poor outcome was defined as 3, 4, or 5. All the patients in follow-up were assessed by 3 senior consultants in follow-up.

RESULTS

Patients in both groups were comparable in age of presentation and length of aganglionic segment and the difference between the two groups was statistically significant. The most common presentation was abdominal distension followed by inability to pass meconium, then enterocolitis and constipation and lastly bilious vomiting.

The average operating time for DP was 161 min. (range 140-180 min.) while for TEPT it was 146 min. (range 120-170 min, table 2).

Figure 2

Table 2: Comparison between two the groups

	Group A	Group B
Per-operative time	161 min. (range140-180)	122 min. (range110-150)
Commencement of oral feed	6th-8th day	5th-6th day
Hospital stay	11 days (range 9-15)	8 days (range 6-11)

Peroral feed was started on the 7 th day in group A whereas in group B it was started on the 5 th . Hospital stay for patients in group A was 11 days (range 9-15) whereas in group B it was only 8 days (range 6-11). The difference between the two groups was not found to be statistically significant.

Complications were more common in group B (constipation requiring suppository/enema – 8 patients, enterocolitis – 6, stricture in 6 patients of which two required re-operation and four were managed by regular anal dilatation in group B whereas three patients in group A had constipation and two had enterocolitis, table 3).

Figure 3

Table 3:Complications

	Group A (n=24)	Group B (n=20)
Enterocolitis	2 (8.4%)	8 (40%)
Constipation	3 (12.5%)	8 (40%)
Stricture at anal region	None	6 (30%)#
Requirement of anal dilatation	None	6(30%)
Retained spur	2(8.3%)	None
Recto-vesical fistula	0	1(5%)
Revised surgery or diversion	1	3 (15%)
Frequency of stool at time of discharge	10-12/day	8-11/day
Frequency of stool at 6 month follow-up	5-8/day	4-6/day
Frequency of stool at 15 months	3-4/day	2-4/day
Death	1*	0
Total complications	12/24	28/20

The higher incidence of enterocolitis in group B was found to be statistically significant (p-value = 0.02)

The incidence of stricture at the anal region was statistically higher in group B in comparison to group A (p-value = 0.01)

one of these required diverting colostomy and one required

stricturoplasty.

* due to associated congenital cardiac disease.

Incidence of enterocolitis and anal stricture was significantly higher in group B in comparison to group A. P-value was 0.02 for enterocolitis and 0.01 for anal stricture (table 3).

In follow-up, patients of both groups were continent with minimal soiling of the clothes (the frequency of stool and soiling of clothes was higher in group B during initial follow-up); the patients in group B required anal dilatation (due to stricture formation) whereas patients of group A were passing stool properly. Satisfactory results according to the scoring system were found in 17 (70.3%) and 10 (50%) patients of group A and B, respectively.

DISCUSSION

A number of operative strategies have been described for Hirschsprung disease. All are perceived to have relative merits and weaknesses, supported in some cases by medium and long-term outcome data [7,8]. One of the essential limitations of an entirely TEPT is the proximal extension of the aganglionic segment beyond the sigmoid colon; so we have included cases with short aganglionic length, that is diseases with transitional zones at the recto-sigmoid or distal to it, though a long segment or unclear transition zone should not be considered an absolute contraindication for TEPT approach. The procedure can be started transanally, switching to an additional laparatomy if the transition zone cannot be reached from below [8]. The remaining seromuscular cuff after conventional EPT or TEPT has been accused for the development of postoperative obstructive symptoms, constipation, and enterocolitis in some patients [3]. In the present study, the transition zone was confirmed by frozen section biopsy. Albanese et al. [9] left a considerable long cuff, but they cut it posteriorly. One of the crucial points of criticism for the TEPT approach is the significant stretching of the anal sphincters during mucosectomy with its potential impact on postoperative continence status particularly in older children with marked hypertrophy and dilatation of the colon [8,10]. This may be the cause of transient soiling and increased frequency of bowel motions in patients of group B (probably because of the stretch effect). Another critical issue is related to the relatively distal level of rectoanal anastomosis in many patients undergoing TEPT technique.

A low anastomosis at or distal to the dentate line may damage the delicate nerve endings that play a part in anorectal continence [8]. Patients with lower anastomosis at or distal to the dentate line were associated with higher frequency of transient soiling for more prolonged periods than those with more proximally located anastomoses as seen in our follow-up patients.

Enterocolitis has been considered one of the main problems in patients with HD both before and after definitive treatment. The incidence of enterocolitis in the present series was 25% which is comparable to other reported series [13]. Enterocolitis was found to be significantly higher in group B (40%) in comparison to 8.4% in group A (p=0.02) and is similar to other reported series [3,10,11,12] whereas Mohamed et al. [14] reported lower incidence of enterocolitis in patients operated by TEPT procedure.

Stricture formation is common in TEPT technique; there is a general tendency to reserve anal dilation or bouginage to cases with existing or potential risk of stricture formation [6]. In the present series, anal stricture was observed in 8 (40%) cases in group B, whereas none of the cases of group A developed anal stricture (table 3). We believe that postoperative routine anorectal bouginage is an effective tool to prevent the occurrence of anal stricture and to decrease both the frequency as well as the severity of enterocolitis, particularly in neonates and young infants.

Constipation was observed in 27.7% (12/44) in the present series which was slightly higher than in other reported series [15,16]. The incidence in the literature ranges from 6% to 27%. The incidence of constipation was higher in group B (40%) in comparison to group A. (12.5%), though this was not found to be statistically significant. The results in the present series were contrary to the results of Mohamed et al. [14]

Functional results were slightly better in group A in comparison to group B, but were not found to be statistically significant in long-term follow-up; satisfactory results were found in 70.3% of cases in group A and only 50% in group B. The main objective of the present study was to compare the results of single-stage DP and single-stage TEPT; so we have not included the patients with long aganglionic segment of bowel. Both groups were identical in age, sex and aganglionic length of the bowel (table 1) and all cases were operated by the same surgeon to avoid any discrepancy in the results.

CONCLUSION

Single stage Duhamel pull-through operation is a safe procedure in neonates with satisfactory results. TEPT is also

a good alternative, but the complication rates are higher.

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