

## Evaluation of the effect of one-stage transanal endorectal pull-through (TERPT) technique on defecation in patients with Hirschsprung's Disease over the last 10 years in Tehran Pediatrics Medical Center

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### Abstract

**Introduction:** One-stage Transanal Endorectal Pull-Through (TERPT) is one of the methods used for the treatment of Hirschsprung's disease. The purpose of our study was to evaluate complications of surgery specially does related to defecation in patients undergoing pull-through surgery in Tehran Pediatrics Medical Center during the past 10 years.

**Materials and Methods:** This descriptive study was performed on patients undergoing one-stage TERPT operation in Tehran Pediatrics Medical Center over the past 10 years.

Exclusion Criteria included: the parent's lack of consent to participate in the study, age below 3 years and inability to complete the checklist. For all patients, a check list was completed that included: Age, family history of the disease, readmission, follow-up period, presence of soiling, complete or partial fecal incontinence, constipation, use of laxative, use of boogie, presence or absence of RectoAnal Inhibitory Reflex (RAIR) in manometers, stricture in rectal examination and enterocolitis before and after surgery. Data analysis was performed using SPSS software version 19.

**Results:** Our patients were 4.81% male and 3.9% of them had an underlying disease, such as heart or kidney disease. Only 6.81% of them

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## Keywords

- Hirschsprung's Disease
- Transanal endorectal pull-through (TERPT)
- Soiling
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- Constipation

had to be re-admitted due to enterocolitis. Soiling was found in 5.39% of patients, complete and partial fecal incontinence were noted in 3.2% and 7.4% of them respectively. The incidence of fecal incontinence was 7%. Constipation occurred in 6.11% of cases and 3.9% of cases did not have RAIR in manometry. The incidence of rectal stenosis was 7% and 93% of patients had transitional zone (TZ) in barium enema before the operation. The result showed that there was no significant relationship between post-operative complications in both sexes ( $p < 0.05$ ).

**Conclusion:** This study shows that the complications of Endorectal Transanal pull-through surgery as a method for treatment of Hirschsprung disease are infrequent. Therefore, this method can be considered as a selective method. Further investigations are recommended to prove this matter.

## Introduction

Hirschsprung's disease is a congenital disorder in which parts of the large and small intestinal wall are ganglion free. Neural cells or ganglion cells migrate from proximal to distal and from the outside of the intestine into the intestine during the embryonic period<sup>1</sup>.

Normally, these neurons are located throughout the gastrointestinal tract, in mucosal and muscular layers. In Hirschsprung's disease, the process of migration has not finalized in the fetus. Therefore, depending on when this occurs in the embryonic life, some parts of the distal intestinal tract will not have ganglion cells.

Presence of ganglion cells is essential for normal intestinal peristalsis and absence of ganglion cells impairs bowel peristalsis, leading to maldigestion and malabsorptive conditions. Symptoms of this disease are: delayed meconium passage, abdominal distention, and intolerance of oral intake, biliary

vomiting during neonatal period, constipation, abdominal distension, growth retardation and enterocolitis at higher ages<sup>2,3</sup>.

The only way to handle this disease is surgery by using various techniques. In different surgical techniques, affected parts which lack ganglions are resected and the healthy proximal piece with ganglions are anastomosed above the dentate line. In the past, this was done in 2 stages of surgery. In the first stage, the proximal ganglion containing part was exteriorized as a stoma, and in the second stage, the distal part of stoma that was ganglion free was resected. Then the healthy part or ganglionic part was anastomosed above the dentate line<sup>4</sup>.

Currently this technique is being used in tertiary centers (in specific populations). To perform the second stage, there are several techniques that can be used such as Swenson and Duhamel and soave method. In recent decades, one-step Trans Endorectal pull-through has been proposed. This method

has advantages over previous methods. One of the benefits is being a one-step operation. There is no need for colostomy and subsequent laparotomy. On the other hand, some studies point out the other fact that surgery with anal manipulation may cause some degree of stool incontinence in the future. At the pediatric Medical center, transanal endo rectal pull-through surgery has been done since 10 years ago. TREPT is a surgical technique that has been used to treat patients with Hirschsprung's disease in recent years. Some researchers believe that dilation during surgery and extensive manipulation by the surgeon in anal canal leads to anal sphincter injury that can lead to some degree of incontinence in the future<sup>5-6</sup> although, some studies denied this theory. In a study conducted in Norway in 2010; researchers found differences between patients who underwent TRERP versus LEPT (laparotomy abdominal perineal pull-through) in stool continence<sup>1</sup>. In a study in 2011, comparing patients underwent TREPT (soave) versus transabdominal (soave) showed that there was no statically significant difference between the 2 groups regarding stool continence. In this study, patients with low inappropriate fecal continence score rate (FCSR) had no clear impairment on pelvic floor musculature and their sphincter complex according to MRI and EMG studies<sup>2</sup>.

The aim of this study is to investigate the degree of fecal incontinency and complications of patients undergoing a one-stage TERPT over a 10 year period.

## Material and Methods

This study investigated patients with Hirschsprung's disease over the last ten years in Children's Medical

Center who underwent one-stage TERPT.

All participants were evaluated regarding stool continence with a questionnaire. All parents who participated in filling the questionnaire were assured in terms of confidentiality of their children's information.

We identified patients who were treated using the TERPT method over the last 10 years in our hospital. Then a manometry was performed for all patients. Also patients with other underlying disease and patients whose parents refused to do manometry were excluded from this study. Also all patients who underwent TREPT but pathology didn't find proof of Hirsch sprung's disease were excluded. During physical examination, rectal examination was performed for all patients. The questionnaire was completed by a pediatric surgeon and anorectal manometries were reported by a pediatric gastroenterologist.

After completing the checklist, data were analyzed using SPSS software version 19. Descriptive statistics including frequency tables, plotting and central indicators and dispersion were used. Also one-way chi square and ANOVA test was used.

## Results

In this study of 80 patients with Hirschsprung's disease who underwent TERPT surgery, only 43 patients fulfilled our inclusion criteria. Exclusions were made due to:

- 1) All children less than 2 years were excluded since our manometry device could only be used on older children.
- 2) Patients who had poor bowel prep.

3) Patients who failed to attend post operation visit.

Demographic characteristics of our patients are presented in **Table 1**. It should be noted that none of our cases needed reoperation. Some post-surgical complication after TERPT and their method of treatment have been shown in **Table 2 and 3**.

Boogies were used for 14 cases during a mean time of  $7.77 \pm 8.9$  months. The minimum time of use was 1 month and the max was 24. The average boogie diameter used was  $14.35 \pm 1.82$ , the minimum size was 12 and the maximum diameter was 16. The incidence of enterocolitis before and after surgery

**Table 1-** Demographic characteristics of children with Hirschsprung's disease who had undergone one-stage (TERPT)

| Variables  | Situation | Number            | Percentage |
|--|-----------|-------------------|------------|
| Sex  | Male      | 35                | 81.4       |
|  | Female    | 8                 | 18.6       |
| Underlying Disease   | Positive  | 4                 | 9.3        |
|  | Negative  | 39                | 90.7       |
| Family History of disease  | Positive  | 4                 | 9.3        |
|  | Negative  | 39                | 90.7       |
| Re-admission   | Positive  | 8                 | 18.6       |
|  | Negative  | 35                | 81.4       |
| Age of patients at the time of surgery (Month, Mean $\pm$ SD)    |           | 5.12 $\pm$ 5.37   |            |
| Age of patients at the time of our study (Month) (Mean $\pm$ SD) |           | 17.33 $\pm$ 70.9  |            |
| The duration of follow-up (Month) (Mean $\pm$ SD)                |           | 17.03 $\pm$ 65.53 |            |

**Table 2-** Fecal incontinency in patients who had undergone TERPT in Tehran children's Medical center over a decade

| Variable                      | Situation | Number | Percentage |
|-------------------------------|-----------|--------|------------|
| Fecal Soiling                 | Positive  | 17     | 39.5       |
|                               | Negative  | 26     | 60.5       |
| Complete Fecal incontinency   | Positive  | 1      | 2.3        |
|                               | Negative  | 42     | 97.7       |
| Incomplete Fecal Incontinency | Positive  | 2      | 4.7        |
|                               | Negative  | 41     | 95.3       |

**Table 3-** Complications other than fecal incontinency and their post-surgical treatment methods in our study

| Variables                              | Situation | Number | Percentage |
|--|-----------|--------|------------|
| Constipation                           | Positive  | 5      | 11.6       |
|  | Negative  | 38     | 88.4       |
| Laxative consumption                   | Positive  | 2      | 4.7        |
|  | Negative  | 41     | 95.3       |
| Use of boogie                          | Positive  | 14     | 32.6       |
|  | Negative  | 29     | 67.4       |
| RAIR in manometry post operation       | Positive  | 39     | 90.7       |
|  | Negative  | 4      | 9.3        |
| Stricture in rectal examination        | Positive  | 3      | 7          |
|  | Negative  | 40     | 93         |
| Tranzitional Zone in pre-op evaluation | Positive  | 40     | 93         |
|  | Negative  | 3      | 7          |

**Table 4-** Relationship between enterocolitis before and after surgery in our patients (P-value=0.04)

| Enterocolitis before and after surgery | Enterocolitis before surgery |            | Enterocolitis after surgery |            | Relation between Enterocolitis before and after surgery |            |
|--|------------------------------|------------|-----------------------------|------------|---|------------|
|  | Number                       | Percentage | Number                      | Percentage | Number  | Percentage |
| Non affected                           | 40                           | 93         | 37                          | 86         | 4   | 67         |
| Affected                               | 3                            | 7          | 6                           | 14         | 2   | 33         |
| Total                                  | 43                           | 100        | 43                          | 100        | 6   | 100        |

is shown in **Table 4**. Analyzing the data using fisher's exact test shows that there is no significant relationship between post-operative complications in patients whom underwent TERPT surgery and their gender. ( $P>0.05$ )

## Discussion

In this study, a total of 43 patients were studied from a total of 80 patients with hirschsprung's disease. Constipation can occur several months after surgery and may depend on the type of operation. This complication has been reported in both Duhamel

and Soave method more than other methods<sup>3</sup>. In general, incidence of constipation is 11.2%. Fecal urgency is seen immediately after TERPT, up to 10 times a day. Of course, this Figureure is also seen in the first trimester after surgery and during the next 6 months it will decline to 2 to 3 times a day.

Generally, constipation after TERPT is a common compliant without histopathology linkages and can be treated conservatively. The rate of constipation in various sources is reported to be between 6 and 30%. PURI and NIXON reported constipation in

34.5% of children after surgery<sup>4</sup>. In another study 27% of children had constipation which has been resolved with medical treatment<sup>5</sup>.

The incidence of enterocolitis in different studies varies between 20 and 40%. In another study, the incidence of Enterocolitis was 10% which increased up to 46%<sup>6</sup>. In our study the incidence was 14%.

The incidence after surgery was higher in the first 2 years. Some surgeons recommended rectal washing to prevent post-operative enterocolitis. Some other surgeons recommended oral intake of metronidazole for 2 month after surgery<sup>3</sup>.

The incidence of stenosis has been reported after one-step TREPT to be up to 20% that often responds to dilatation.

Patients who have had early leakage after surgery are more likely to suffer from stenosis and enterocolitis is even more common in these patients<sup>3</sup>.

In our study 7% had anastomotic stenosis and 16% of post-operative enterocolitis had anal stenosis.

None of the complications of Hirschsprung's surgery will affect the patient's quality of life as severely as fecal incontinence can. The overall rate of incontinency reported is 3 to 8% and usually as the baby grows-up the incontinency will improve.

We recommend medical treatment for constipation during this period and if it doesn't respond surgery may be perform<sup>3</sup>.

The incidence rate of incontinency in adults undergoing abdominal TERPT surgery is higher

but we couldn't find any statistics regarding pediatric patients.

It should be mentioned that incontinency without constipation is curable with biofeedback. Over the time when incontinency is treated, about 90% of patients will get normal bowel function.

The incidence of soiling after TERPT is 2.3 because of 1) sensory abnormalities in return 2) sphincter dysfunction and 3) Incontinency.

According to other studies the most common cause of soiling after TERPT is false incontinency. We can prescribe Loperamide if it is due to hyper peristalsis. It is of note that our study was conducted in the early post operative period, thus it may be better to reassess children with fecal incontinency with EMG and MRI in the future.

Fecal incontinency will affect personal life the fear of incontinency can lead of significant reduction in social activities. Because of that it is important to determine quality of life in patients in addition to fecal continence. TERPT method has been used for the last 2 decades, and it is becoming more common in treatment of Hirschsprung's disease every day. To obtain better results it is recommended to be done at an earlier age<sup>7</sup>.

In contrast with the transabdominal method TERPT is less invasive and post-operative complication and pain is much lower<sup>8</sup>. Postoperative results from TERPT have not yet been evaluated satisfactorily with EMG. In addition, there are no large volume studies that could show the incidence of incontinence after this surgery<sup>9-11</sup>. In our study EMG for evaluating the incidence of incontinency was not performed<sup>12, 13</sup>.



## Conclusion

Although our study showed that the TERPT technique results in good long term results but

further examination for fecal incontinence, quality of life and manometric studies are required to achieve a definitive conclusion.

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