

Interest of Anorectal Manometry in Long-Term Follow-Up of HIRSCHSPRUNG Disease Patients who Underwent Transanal Pull-Through Procedure

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DOI: 10.29322/IJSRP.12.06.2022.p12633
<http://dx.doi.org/10.29322/IJSRP.12.06.2022.p12633>

Paper Received Date: 19th May 2022
Paper Acceptance Date: 4th June 2022
Paper Publication Date: 14th June 2022

Abstract- Objective:

determine the role of Anorectal Manometry in the long-term follow-up of HIRSCHSPRUNG disease patients who undergo Transanal endorectal pull-through procedure.

Methods:

the study included patients who were operated on for HIRSCHSPRUNG disease using Transanal pull-through surgical technic, after 6 months or more of the surgery.

Anorectal manometry (ARM) was performed; clinical characteristics and their postoperation bowel function were collected.

Study type:

Comparative retrospective study

Level of evidence:

Level VI

Results:

30 patients were enrolled. 14 patients were incontinent. Anal Resting pressure of incontinent patients was significantly lower than continent patients (32.46 ± 8.6 mmHg versus 45.23 ± 9.1 mmHg; $P < 0.05$, t-test).

Four patients (3 continence and 1 incontinence) have a positive RAIR, which is of no statistical value.

Conclusion:

AM could be an objective method for investigating postoperative complications and provides useful findings in patients presenting with defecation disorders, and AM may help in decision making more objectively than just clinical examination.

Index Terms- Follow-up studies; HIRSCHSPRUNG Disease; Manometry; defecation disorders

I. INTRODUCTION

There are many recent advances related to surgical techniques for HIRSCHSPRUNG disease (HD). However, postoperative outcomes remain variable [1].

The Transanal endorectal pull-through (TERPT) was reported by De La Torre- Mondragon in 1998[2] and has been widely performed because it has a low degree of invasiveness[3], and today abdominal access is considered to be necessary only in selected cases [4].

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<http://dx.doi.org/10.29322/IJSRP.12.06.2022.p12633>

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Many children with HD have a good outcome from surgery, but long-term follow-up studies have shown ongoing problems in some of the children with fecal incontinence and constipation being the most prominent symptoms [5].

Post-operative anorectal manometric evaluation of the patients after surgery gives information for the function of the anal canal and rectum[6]but is still not routinely used for long-term follow-up.

II. METHODS

1.1. study design:

a comparative retrospective study was conducted at Tishreen University Hospital in Latakia, Syria. In addition, was approved by the research ethics committee of Tishreen University.

Was performed with a (DYNO SMART-SN-ZC0035-IP 20 0476) device, connected to (HP) computer. Data analyzed by (DYNO-3000) software.

The catheter was a 3mm (9Fr) water perfused catheter, with a central canal of 1.1mm opens into a balloon of 60ml in volume, has four lumens of 0.8mm arranged helically along the distal part of the catheter at a distance of 0.5cm.

1.2. Patients:

The study included patients who were operated on for HIRSCHSPRUNG disease (confirmed with rectal biopsy) in Tishreen University Hospital using Transanal pull- through surgical technic, and 6 months or more have passed on the surgery.

The overall number of children with trisomy 21 in our who had HD IN OUR facility were significantly high, but we didn't have any in our group study.

Demographic and clinical data were collected; gender, date of birth, age at the first symptom arise, age at the surgery, enterostomy, length of resected colon segment, operation complications, and postoperative bowel movement complications. patients were investigated and evaluated for the presence of constipation (according to ROME IV criteria) and fecal incontinence (according to Wingspread classification).

Anorectal manometry (ARM) was performed on all patients and finding were collected to analysis containing resting anal pressure (RP) and Rectoanal inhibitory reflex (RAIR).

All of the patients were followed up on a regular base and all were undergo a postoperation anal dilation starting 3 weeks postoperatively twice a week until 6 months after the surgery.

1.3. Assessment of short-term outcomes:

All patients were asked for postoperative complications, and were included; postoperative enterocolitis, anastomotic stricture, leakage, colonic torsion, intestinal obstruction, or none. In addition, all complications were assessed approximately 3 months postoperatively.

1.4. Assessment of long-term outcomes:

Long-term problems in children with HD include ongoing obstructive symptoms, soiling, and enterocolitis. It is important for the surgeon to follow these children closely, at least until they are through the toilet training process, in order to identify and provide timely treatment for these problems.

1.4.1 Fecal incontinence:

Patients were divided into four groups according to **Wingspread** classification [1]; 1-Excellent (totally continent, toilet trained with no medication needed).

2- Good (rarely soiling, except during stressful exercise). 3-fair (intermittent fecal soiling, urge incontinence).

4- Poor (constant soiling or smearing).

Patients are classified as continent (excellent, good) or incontinent (fair, poor).

Fecal continence was considered to have been acquired normally in children at age of 4 years and older according to Rome IV criteria for fecal incontinence [7].

1.4.2 Constipation:

Patients divided into two groups according to **Rome IV** criteria [8] for constipation: Even if they have constipation or do not.

By investigating the stooling frequency, the use of laxatives und/or enema.

A patient was considered as suffering from constipation if defecation was only possible with laxatives, or occurred less than 3 times per week.

1.4.3 Enterocolitis:

The etiology of HAEC is unknown and is probably multifactorial. Stasis caused by functional obstruction permits bacterial overgrowth with secondary infection.

Infectious agents such as *Clostridium difficile* or rotavirus have been postulated as being causative, but there are little data to support a specific pathogen.

Enterocolitis may be present both before and after operative correction, and can range in severity from mild to life threatening. HAEC is more common in younger children.

1.5 Anorectal manometry test:

1.5.1 Anal resting pressure (RP):

Was Identified for all patients, at beginning of the test, a minimum 5-minute familiarization period was undertaken. Then the anal resting pressure was recorded in mmHg.

1.5.2 Rectoanal inhibitory reflux (RAIR):

RAIR was investigated with inflating intrarectal balloon from 5ml to 60ml, the reflux considered present if there was fallen $\geq 20\%$ of RP and last for 5sec at least.

1.6 Statistical analysis:

Data were reported as mean and standard deviation or median and range For continuous variables, according to their distribution.

The chi-square test or the Fischer exact test was used to compare proportions. The independent t-test was used to compare means, and all tests were two-sided. A P-value < 0.05 was considered significant.

We use IBM SPSS Statistics (version 20) for statistical analysis.

III. RESULTS

30 patients were enrolled in our study and were 4 years old and older, we exclude 7 patients due to their age (less than 4 years because we couldn't evaluate the continence status)

Table.1 shows the number and the classification of the 30 children who performed ARM and were at the age to assess their fecal status according to Wingspread classification and showed that half of them were in the continence group(excellent and good) (53.3%).

Continence scoring according to Wingspread classification	N(30)	percentage %
Excellent	7	23.3%
good	9	30%
Fair	9	30%
poor	5	16.7%

Table(1): classification of the 30 children who performed ARM according to Wingspread classification

[Table.2] demonstrate the characteristic differences between the two group (continence and incontinence), the mean age of (5.89 \pm 2.4 y) for the continence group and (5.53 \pm 1.7 y) for the incontinence group with a p-value of 0.7

Our patients were divided into 18 males (11 continence and 7 incontinence) and 12 females (5 continence and 7 incontinence) with a p-value of 0.2, the age of the first presentation were also with no statistical differences, the mean age in the continence group was (0.63 \pm 1.07 y) and in incontinence was (0.62 \pm 1.2 y) with a p-value of 0.9

Mean age at the operation was (1.98 \pm 1.2 y) in the continence group and (2.79 \pm 2.8 y) in incontinence with a p-value of 0.3

Mean length of the resected colon segment were (34.93 \pm 10.1cm) incontinence and (39.14 \pm 16.5cm) in incontinence, p-value 0.4

Patients variables	N	Continence	Incontinence	P-value
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SEX n(n/N,%)	18			0.2
male		11(68.8%)	7(50%)	
female	12	5(31.3%)	7(50%)	
AGE (years)		5.89 ± 2.4	5.53±1.7	0.7
Age at first presentation(years)		0.63±1.07	0.62±1.2	0.9
Age at the operation (years)		1.98±1.2	2.79±2.8	0.3
Length of resected colon segment (cm)		34.93±10.1	39.14±16.5	0.4
Enterostomy n(n/N,%) Yes No	6 24	4(25%)	2(14.3%)	0.4
Enterocolitis n(n/N,%) Yes No	1 29	0(0%)	1(7.1%)	0.5
Anastomosis leakage n(n/N,%) YES NO	3 27	2(12.5%)	1(7.1%)	
Constipation n(n/N,%) Yes No	2 28	2(12.5%) 14(87.5%)	0(0%) 14(100%)	0.1

Table(2): the characteristic differences between the two group

6 out of the 30 were had an Enterostomy before the corrected operation (4 continence and 2 incontinence) p-value 0.4

Post-operation complications were appeared in 4 out of 30 (3 anastomosis leakage and 1 enterocolitis) with no statistical differences.

The presence of constipation was found in two patients who were incontinence group with a p-value of 0.1

We notice that there were not any statistical differences between the two groups in the presence of constipation, postoperative complication, enterostomy, the length of the resected colon, and demographic changes.

The manometric finding of continent patients compared with incontinent patients shown in [table.3] we can see that the Anal Resting pressure of incontinent patients was significantly lower than continent patients (32.46 ± 8.6 mmHg versus 45.23 ± 9.1 mmHg; P < 0.05, t-test) and four out of 30 patients (1.2%) showed a relaxation reflex what was considered as a reappearance of RAIR (3 incontinence group an 1 in incontinence group) p>0.05 with no statistical differences.

Manometric findings	N	CONTINANCE	INCONTINANCE	P-value
RAIR n(n/N,%)				
Yes	4	3(18.8%)	1(7.1%)	0.3
No	26	13(81.3%)	13(92.9%)	
Anal resting pressure (mean±SD,mmHg)		45.23 ± 9.1	32.46 ± 8.6	0.001

Table(3): The manometric finding of continent patients compared with incontinent patients

IV. DISCUSSION

TERPT is a worldwide popular HD surgical technique [3.9], which needs no abdominal opening besides the low risk of adhesion, the pleasant cosmetic results, less postoperative pain, and low complications [2.3].

Review of the literature showed that all technical procedures available for HD patients (TERPT included) are accompanied by certain complications [10]

In our study, all the patients were undergone TERPT so we did not compare the complications of a different technique.

Performing single-stage TERPT in the non-neonatal period though to be more appropriate than the neonatal period because of the high rates of perianal excoriation, anastomotic stricture or leakage, postoperative enterocolitis, and incomplete continence postoperatively in neonates than nonneonates[11], our group study has a large variety of age at the time of the procedure with a median age of (2.36y) and that put a big question in front of us about the optimal operation age.

The short-term postoperation complications like anastomosis leakage and enterocolitis has been decreased worldwide due to the development of surgical equipment, colonic preoperation preparation, and increased surgical expertise [3.12.13]

Enterocolitis has been considered one of the main complications in patients with HD, but the rates vary [14], a very small number of our patients have complications after the surgery (3 had an anastomosis leakage and 1 had enterocolitis).

The relatively low incidence of enterocolitis in our series may be due to the combined short the V-shaped partial resection of the muscular cuff and our routine postoperative anal dilatation protocol aside with the high experiences in performing TERPT over those years and by the same consideration other complications such as anastomotic stricture, constipation, and spasm of the internal sphincters may also be decreased for the same reasons as we show in [table .2]

On the other hand, the long-term complications still elevated with the defecation disorders being the most prominent [1.15] and all the reports around the world

Demonstrate that the results of these disorders are not always satisfactory after this operation [16]

We exclude seven children from our study because they were younger than 4 years that we could not evaluate the continence (according to Rome IV criteria)

The remaining 30 children were distributed in four categories depending on the parents' notes for defecation habits, there were 53.3% of them in the category excellent and good (Wingspread classification) and considered their continence.

The main target in our study was to determine the differences between the two groups continence and incontinence; we study all the demographic differences for the 30 patients.

Fecal continence depends on three main factors, voluntary sphincter muscles, anal canal sensation, and colonic motility [17]

we didn't see any statistical differences among the two groups in the aspect of age, although we did have a variety of ages from 4 years to 11 years, as well as the sex, the age at the operation, even in the length of colon resected segment we couldn't related to the post-operation continence, and that is a very interesting result that we had a range from 20cm to even a total colectomy.

As well as the postoperation complication had no differences in the continence, and the presence of Enterostomy before the correction surgery has no significant difference.

Constipation, when left untreated, can be extremely incapacitating, and in its most serious forms can produce a form of fecal incontinence known as overflow pseudocontinence[15] and the reasons for constipation and incontinence are myriad, including long muscular sheaths, tissue injury in an inaccurate procedure, and false judgment of intraoperative biopsy [13.18] as well as the uncleanness of the reasons that why some children do well after surgery for HD, and others have not[6], in our study, we didn't find statistical differences in the term of the presence of constipation. Although one must be kept in mind that in some children, the reason for obstructive symptoms may be caused by anal stenosis [6], we exclude the stenosis depending on the anal dilations protocol performed postoperatively and approved by physical examination during the follow-up.

Postoperative outcomes of TERPT have not yet been adequately assessed regarding the postoperative ARM and/or EMG, and There are few reports about ARM during follow-up of children operated on for HD, and that is the value of our study that till now AM role still seems controversial in terms of the uses of different anorectal manometry systems (conventional versus high-resolution anorectal manometry [HRAM]) beside the lack of normal value[19], in our study we use the DYNOSMART-300 manometry system which was the only option we had in our facility.

The anorectal manometry test helps us to understand the essential pathophysiological process that is responsible for the persistent symptoms [6], and its results help us to evaluate compliance of the rectum for storage and resistance of anal sphincter against leakage of rectal contents [5]

Anal dilatation and stretching, which is required to perform a Transanal mucosectomy, has been suggested as a potential source of future functional problems.

However, it has been shown recently that the manometric resting sphincter pressure is comparable between patients operated on transanally and those operated with abdominal endorectal pull-through [20]. However, it is important to investigate the anorectal function in symptomatic children at an early stage to appropriately treat any functional disturbance of the rectum and anus [5]

Resting pressure of the rectum in healthy children assesses the function of the internal sphincter, whereas squeeze pressure is the function of the external sphincter [21]

S. Kumar et al [14] study was aimed to enroll AM on healthy children, the average Mean resting pressure in their subjects was 41.24 ± 12.40 mm Hg (range, 21.3-67.3 mm Hg).

In our study, the mean RP in the incontinence group was very low in comparison with the continence one (32.46 ± 8.6 mmHg Versus 45.23 ± 9.1 mmHg) with a p-value of which is considered of statistical value and it's compatible with what Viet Q Tran, et al [1] reported with

A wide range of mean RP between the continence and incontinence groups (63 ± 11 mmHg versus 47 ± 12 mmHg).

In the literature, several authors stated that patients with true incontinence present a significantly lower resting pressure than normal subjects [22].

There is no agreement with the absence or the presence of RAIR during anorectal manometry after surgical procedures for HD [6] and the re-establishment of RAIR was not clinically correlated with any positive long-term outcome in our cases as well in the other studies [1,23].

Our results show that 4 patients (3 continence and 1 incontinence) have a positive RAIR which is of no statistical value, and that which corresponds to what literature reported.

Our study was enrolled in a pediatric specialist facility, with a purpose was to determine the role of ARM in the long-term follow-up.

Results after TERPT for our HD patients were relatively acceptable, and the Complications such as persistent constipation, enterocolitis, and fecal incontinence were evaluated and studied to compare them with the results we had from ARM.

Our experience shows that ARM could be an objective method for investigating postoperative complications and provides useful findings in patients presenting with defecation disorders, and ARM may help in decision making more objectively than just clinical examination and that was our main aim.

Conflict of interest None.

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