

# Three-dimensional high-resolution anorectal manometry in children after surgery for anorectal disorders

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**Abstract.** – **OBJECTIVE:** Three-dimensional high-resolution anorectal manometry (3DHRAM) is the most precise tool for assessing the function of the anal canal. Our aim was to evaluate children after surgery for anorectal disorders using 3DHRAM.

**PATIENTS AND METHODS:** We prospectively enrolled 43 children (30 males; mean age: 7 years) after surgery for Hirschsprung's disease, anal atresia, or after proctocolectomy. Manometric data were compared to raw data obtained from previously studied children without symptoms arising from the lower gastrointestinal tract. Correlations between manometry and symptoms were evaluated.

**RESULTS:** The lowest values of the resting pressure, squeeze pressure, and pressure of the puborectalis muscle were observed in the anal atresia group (55.6 mmHg, 121.7 mmHg, and 44.17 mmHg, respectively). Compared to asymptomatic children, the lowest mean resting pressures were observed in those with non-retentive fecal incontinence (61.3 mmHg,  $p < 0.000$ ). The receiver operating curve cut-off value for the mean resting pressure between asymptomatic children and incontinent patients was 68.5 mmHg. The thresholds of urge were significantly higher in constipated patients compared to asymptomatic patients (87.5 cm<sup>3</sup> and 30 cm<sup>3</sup>, respectively;  $p = 0.003$ ).

**CONCLUSIONS:** 3DHRAM may be a useful tool for assessing the function of the anorectum of children after surgery (NCT02296008).

*Key Words:*

Anal atresia, Children, Fecal incontinence, High-resolution manometry, Hirschsprung's disease.

## Introduction

Children may present a wide spectrum of symptoms that originate from the gastrointes-

tinal tract after lower gastrointestinal surgery. The most frequent and devastating problem is fecal incontinence (FI)<sup>1</sup>, which may occur in up to one-quarter of patients after undergoing surgery for Hirschsprung's disease (HD)<sup>2</sup>, and in more than half of patients after correction of anorectal malformations (ARM)<sup>3</sup>. Children may also present other symptoms after surgery for ARM due to the complexity of the congenital anomaly, such as urological problems (e.g., urinary incontinence, neurogenic bladder dysfunction, and uropathy)<sup>4</sup> due to the presence of genitourinary defects<sup>5</sup>.

Fecal continence is maintained through a complex system involving several factors, such as stool volume/consistency, rectal compliance; anal sphincter function, sensation, and reflex; neurologic innervation; and cognitive ability<sup>6,7</sup>. Incontinence usually occurs when one or more mechanisms are defective<sup>6,7</sup>. Due to the complexity of this phenomenon, systematic algorithms are used in clinical practice<sup>8</sup>. Guidelines for the management of postoperative soiling in children with Hirschsprung's disease<sup>9</sup> classify symptomatic patients with anatomic or physiologic disruption of their continence mechanisms as having "true fecal incontinence", and inadequate sphincter control and abnormal sensation are noted as key factors that should be evaluated first<sup>8,9</sup>. The most important muscle for determining the resting pressure of the anal canal is the internal anal sphincter (IAS). A poor functional outcome may be correlated with the lack of IAS in severe instances of ARM<sup>10</sup> or its improper resting pressure and/or function in HD<sup>11</sup>. In patients after proctocolectomy, fecal continence may be problematic due to the length of the resected bowel and the type of anastomosis<sup>12</sup>. In some cases, the surgical

technique may compromise the sphincters, and this may potentially result in a decreased resting pressure<sup>13</sup>.

A proper evaluation of all the factors responsible for continence is challenging. Several diagnostic tests are currently used, including anorectal and/or colonic manometry, colonic transit time studies, and ultrasonography. Recently, the advent of three-dimensional high-resolution anorectal manometry (3DHRAM) has allowed for the evaluation of anorectal function in a very detailed manner<sup>14</sup>. The method has the potential ability to record the pressure contribution of the different components of the anal canal<sup>15</sup> and detect discrete pressure defects<sup>16</sup> that may have an impact on continence function.

We hypothesize that this manometric technique will allow for a more detailed identification of key mechanisms of fecal continence in these children. Our aim was to unravel the extent of functional disruption of sphincters and sensation with the most advanced manometric technique.

## Patients and Methods

### Study Patients

Children who had undergone surgery for anorectal disorders were prospectively enrolled and underwent manometric evaluation at the Department of Pediatric Gastroenterology and Nutrition at the Medical University of Warsaw in Poland. All of the patients were divided into three groups as follows: HD – after surgery for Hirschsprung's disease, AA – after surgery for anal atresia, and PC – after proctocolectomy. The indications for proctocolectomy were familial adenomatous polyposis, desmosis coli, and inflammatory bowel disease. The recorded data were compared to the raw data that were obtained in our laboratory from healthy children in previously published studies (the HC group)<sup>17</sup>. To assess the correlation between manometry and symptoms, all of the children from the after-surgery and HC groups were divided into groups with respect to their symptoms as follows: asymptomatic (A), non-retentive fecal incontinent (NRFI), constipated (C), and retentive fecal incontinent (RFI). The groups were established according to the Rome III criteria<sup>18</sup>.

This study was approved by the Ethics Committee of the Medical University of Warsaw, Poland (KB7/2013) and registered at *ClinicalTrials.gov* (NCT02296008). The procedures used in this

study adhere to the tenets of the Declaration of Helsinki. All persons gave their informed consent prior to their inclusion in the study.

### Equipment

Manometry was performed using 3DHRAM (ManoScan 360/3D; Medtronic, Dublin, Ireland). The solid-state, rigid probe (64 mm length and 10.75 mm diameter) consisted of 256 pressure sensors (micro transducers) attached to an amplifier and recorder system, which is connected to the computer. Each sensor is 4 mm long and 2 mm wide. Due to the high number of sensors, the catheter is able to measure pressure longitudinally and circumferentially within the anal canal during rest and voluntary squeeze of the anal sphincters, allowing for evaluation of the strength of the individual muscles. The topography of the anal canal pressure is displayed on the computer using specialized software (ManoScan AR 2.1, Covidien/Medtronic) in live mode, allowing for proper positioning of the probe inside the anal canal.

To assess neural reflexes and sensation, a balloon is attached at the tip of the probe. There is a lumen inside the probe through which air can be administered by a 60-mL syringe into the balloon. This allows for the measurement of the thresholds of sensation and recto-anal inhibitory reflex (RAIR). The balloon is 3.3 cm long and is composed of a non-latex thermoplastic elastomer.

Before each examination, the probe was calibrated over a range of 0-300 mmHg, and an in vivo calibration procedure in 36-38°C water was performed once each week.

### Procedure

No routine bowel preparation was used. If stool was present during the digital rectal investigation, then, a 100 mL saline enema was administered 1 hour before the procedure. The patients were investigated in the lying position. At the beginning of the procedure, the probe was lubricated and inserted into the rectum so that the proximal and distal margins of the anal canal were clearly seen on the computer screen. After an accommodation period of 2 minutes, the following conventional manometric parameters were recorded: resting pressure, squeeze pressure (performed twice), the presence of RAIR, and thresholds of sensation. RAIR was evaluated by rapid inflation and deflation of the balloon with incremental volumes ranging from 10-60 mL. A positive RAIR was defined

as a 25% decrease in the mean resting pressure. This reflex was not investigated in children after surgery for Hirschsprung's disease. Thresholds of sensation were obtained by the continuous administration of air into the balloon, which was performed twice. Squeeze pressure and thresholds of sensation were evaluated only in children who cooperated.

### Data Analysis

After each procedure was finished, all of the recorded data was analyzed by dedicated software (ManoView AR v2.1; Covidien/Medtronic, Dublin, Ireland), which allows for the analysis of conventional manometric parameters. Raw data from 256 sensors were used to analyze a 3D picture of the anal canal. Proximal and distal margins of the high-pressure zone (HPZ) were identified with the aid of the implemented software algorithm separately for the resting and squeeze periods. The anal canal was divided into proximal and distal parts and then into the anterior, posterior, left, and right segments, as previously described<sup>17</sup>. This allowed for the aggregation of the 3D pressure map into 8 segments and for the comparison of these segments with respect to the anal canal length. The resting and squeeze pressures of the puborectalis muscle (PRM) were recorded in segments covering its anatomical location.

### Statistical Analysis

Descriptive statistics are expressed as the median and 5<sup>th</sup> and 95<sup>th</sup> percentiles. The distribution of quantitative variables was tested by the Shapiro-Wilk test of normality. The Mann-Whitney U test was used to test differences between two

continuous variables. ANOVA and the Kruskal-Wallis test were used to compare quantitative variables between 4 groups. The Spearman correlation coefficient was used to test correlations, and the  $\chi^2$ -test was used to compare proportions. Receiver operating curve (ROC) analysis was used to determine the optimal cut-off between asymptomatic children and patients with fecal incontinence. Statistical significance was established as a *p*-value of <0.05. All of the analyses were performed using Statistica 13 (Statsoft, OK, USA).

## Results

Forty-three patients (mean age: 7 years; range 1-16 years; 30 males) were included in the study. Twenty-four children (55.8%) were included after surgery for Hirschsprung's disease (HD), 12 (27.9%) after surgery for anal atresia (AA), and 7 (16.3%) children after proctocolectomy (PC). The HD group consisted of patients who had undergone the Duhamel procedure (8 children; 65.2%), transanal endorectal pull-through (15 children, 34.8%), and modified Rehbein (1 child). The AA group consisted of patients who had undergone correction for imperforate anus with perineal fistula (7 children, 58.3%), rectourethral fistula (4 children, 33.3%), and persistent cloaca (1 girl, 8.3%). The clinical characteristics of the subjects are summarized in Table I.

There were no differences between groups after surgery in regard to the conventional parameters. Significant differences were obtained when compared to the HC group (Table II). In the AA group, the mean resting, and squeeze pressures were lower than those of the HC group.

**Table I.** Clinical characteristics of subjects.

Variable/Group (n)	HD (24)		AA (12)		PC (7)		HC (61)	
	Med	Q1-Q3	Med	Q1-Q3	Med	Q1-Q3	Med	Q1-Q3
Age (months)	61	13-197	70	16-195	166	154-187	112	24-210
Weight (kg)	17.9	9.6-59.6	221	0-54.8	56.8	27.8-62.3	30.6	12.5-77
Height (cm)	109	78-173.5	119	80-169	161	143-177	134.5	89-184
Bowel movements per day	2	0.33-10	2	0.3-6	7	1.5-15	1	0.5-3
Incontinence (n; %)	9 (37.5)		5 (41.7)		6 (85.7)		–	
Constipation (n; %)	3 (12.5)		3 (25)		0 (0)		–	
Constipation with fecal soiling (n, %)	3 (12.5)		4 (33.3)		0 (0)		–	
No symptoms (n; %)	9 (37.5)		0 (0)		1 (14.3)		61 (100)	

HD – Hirschsprung's disease; AA – anal atresia; PC – proctocolectomy; HC – healthy children; Med – median; Q1-Q3 – interquartile range.

**Table II.** Conventional manometric parameters in patients after surgery compared to the control group.

Variable	HD			AA			PC			HC		
	N	Med	5-95 c	N	Med	5-95 c	N	Med	5-95 c	N	Med	5-95 c
Mean resting pressure (mmHg)	24	76.2*	55.6-129.2	12	56.6*	12.7-82.8	7	58.9	34.3-135.4	61	89.2	66.4-123.7
Maximum squeeze pressure (mmHg)	24	178.6	82.4-311.5	11	121.7*	38.2-46.8	7	178	81.5-231.9	58	208.5	124.9-292.2
Anal canal length (cm)	24	2.3	1.5-3.5	12	2.55	1-4	7	2.3	2.2-3.7	61	2.6	1.8-3.8
Minimal rectal compliance (cm <sup>3</sup> /mmHg)	24	0.17	-0.2-0.29	12	0.16	0.12-0.20	7	0.18	0.13-0.27	59	0.15	0.12-0.30
Maximal rectal compliance (cm <sup>3</sup> /mmHg)	24	1.02*	0.25-2.18	12	0.99	0.27-2.05	7	0.7	0.43-1.15	59	0.60	0.23-1.45
RAIR (cm <sup>3</sup> )	–	–	–	6	15	10-60	3	20	10-20	61	10	10-30
First sensation (cm <sup>3</sup> )	10	35	10-120	6	20	10-60	5	10	10-20	56	20	10-50
Urge (cm <sup>3</sup> )	10	60	30-179	5	35	20-115	5	50	30-60	56	30	10-130
Discomfort (cm <sup>3</sup> )	10	60	40-210	5	120	50-165	5	60	40-140	56	85	20-180

Med – median; pc – percentile; RAIR – recto-anal inhibitory reflex; HD – Hirschsprung's disease; AA – anal atresia; PC – proctocolectomy; HC – healthy children; \* $p < 0.05$  compared to HC group.

Based on the analysis of segmental pressures, only one segment (distal posterior) differed between the HD and AA groups ( $p=0.03$ ). When segments of the anal canal were compared, it appeared that in the AA group, almost all of the segments had significantly lower values than in the HC group. Only 3 segments in the PC group had lower pressures than in the HC group, and the difference was seen only in the resting state. All segments and pressures of PRM are summarized in Table III.

Conventional parameters in regard to bowel function are summarized in Table IV. Compared to the A group, patients with NRFI and RFI had a significantly lower mean resting pressure ( $p<0.000$  and  $p<0.000$ , respectively). Significantly lower squeeze pressure characterized patients with NRFI and RFI compared to the A group ( $p=0.03$  and  $p=0.008$ , respectively). The ROC cut-off value for the mean resting pressure between asymptomatic children and patients with fecal incontinence was 68.5 mmHg (Figure 1). Similar to the mean resting pressure, a lower resting pressure of PRM in the NRFI group and a lower squeeze pressure of PRM in the RFI group were observed compared to the A group (Figure 2).

The threshold of urge was significantly higher in the C group compared to the A group ( $p=0.003$ ). There was a tendency for a higher threshold of sensation and discomfort to occur in constipated patients and for a lower threshold of discomfort to occur in the NRFI group, but the differences did not reach statistical significance.

The NRFI group was characterized by the highest number of segments with a significantly lower resting pressure compared to the asymptomatic group. During a squeeze, the highest number of segments with a lower pressure was observed in the RFI group. Pressures for all of the segments in regard to bowel function are summarized in Table V.

The mean duration of the manometric procedure was 15:55 minutes (range: 5-27 minutes). The 3DHRAM test was well tolerated, and no adverse effects of the procedure were observed.

## Discussion

Protracted, bothersome symptoms in patients after lower GI tract surgery are a devastating problem often observed in children during long-term follow-up care. The most frequent symptoms are constipation and fecal incontinence,

observed in more than half of all patients<sup>3</sup>. Depending on the complexity of the disorder, children after surgery may also suffer from symptoms arising from improper anatomy or function of the genitourinary tract. Urinary incontinence is found to be present in long-term follow-up care (at least 10 years after correction) with very high prevalence (up to 30.5%) and persists into adulthood<sup>4</sup>.

In summary, our data show that patients after lower gastrointestinal surgery may have a significantly decreased mean resting pressure and/or squeeze pressure. Moreover, these patients may have different numbers of segmental pressure gaps. Decreased pressures are observed more often in patients with fecal incontinence. This is the first study evaluating children after surgery that compares results with normative data obtained from the pediatric population.

In the literature<sup>19</sup>, the role of manometry in the evaluation of children after surgical procedures has been questioned because results are inconsistent. This may reflect the diversity in the types of manometric equipment or different samples pertaining to size and indications for surgery.

Inconsistent data mostly pertains to pressures obtained in studies<sup>20-22</sup> evaluating children after surgery for HD. Some authors reported lower anal pressures in children, consistent with our results, while others found no difference<sup>23-26</sup> or even higher pressures<sup>27</sup> compared to normal values that were adequate for the type of equipment used. As noted above, this discrepancy may be due to the small number of patients who were evaluated or the diversity of the surgical techniques. Different results may also reflect the different lengths of follow-up care. Hsu et al<sup>28</sup> reported a lower resting anal pressure soon after surgery and its increase over time. However, in contrast to this observation, others<sup>29,30</sup> did not report improvement in the low-pressure level many years after the operation.

Data on the morphology of the anal canal after surgery for HD are scarce. Zhang et al<sup>31</sup> found asymmetry in patients with fecal incontinence after the transanal pull-through procedure, which may reflect pressure gaps in the anal canal, which were observed in our study. There is only one study using 3DHRAM for the evaluation of patients after surgery for HD in which the mean resting and squeeze pressures were similar to those obtained in the present study, but in that study the evaluation of the 3D pressure map was made without segmental differentiation<sup>32</sup>. This



**Table III.** The pressures of segments in patients after surgery compared to the control group.

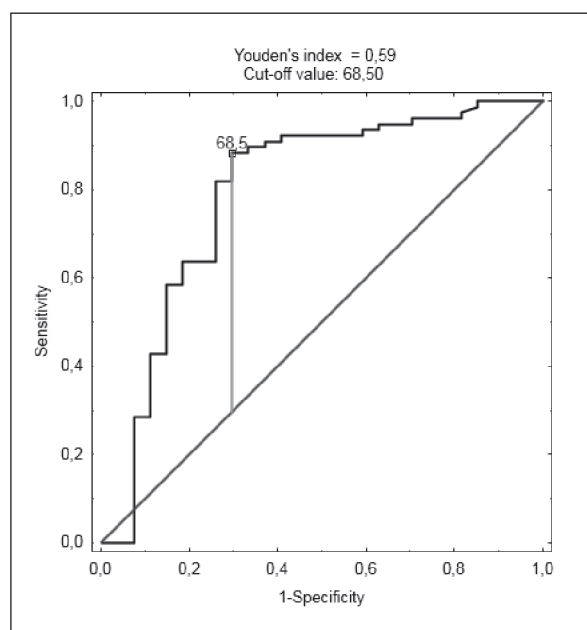
Segment	HD			AA			PC			HC		
	N	Med	5-95 pc	N	Med	5-95 pc	N	Med	5-95 pc	N	Med	5-95 pc
<b>Rest</b>												
Distal anterior	24	42.1*	25-83	12	26.7*	0,7-76	7	29.5*	18-58	61	70.6	44-101
Proximal anterior	24	36.4	21-59	12	27*	5-50	7	30.6	17-72	61	47.2	15-68
Distal posterior	24	36.4	22-73	12	21.6*	-3-47	7	26.7	8-59	61	44.3	18-73
Proximal posteriori	24	47.3*	25-96	12	36.6*	9-70	7	39.2*	21-89	61	71.5	48-97
Distal left	24	63*	41-96	12	45.5*	14-102	7	59.9*	28-84	61	81.6	61-114
Proximal left	24	53.5*	35-109	12	46.2*	10-72	7	59.2	34-73	61	66.2	45-95
Distal right	24	61.8	36-97	12	38.7*	15-78	7	62.1	18-105	61	85.7	60-117
Proximal right	24	52.2	35-106	12	38.2*	14-67	7	53.5	15-77	61	66.7	42-89
<b>Squeeze</b>												
Distal anterior	24	75.6*	30-136	12	41.2*	11-193	7	85.5	48-143	58	135.8	63-197
Proximal anterior	24	51.3	14-113	12	30.3*	7-80	7	60.1	34-85	58	62.3	13-119
Distal posterior	24	69.6	32-128	12	43.3	5-80	7	54.2	39-129	58	59.8	25-129
Proximal posteriori	24	85.7*	42-159	12	64*	10-167	7	62.9	38-212	58	134.4	71-219
Distal left	24	101.3*	62-151	12	76.2*	40-207	7	109.8	60-160	58	130.8	76-188
Proximal left	24	80.8*	38-155	12	62.7*	19-122	7	94.8	49-164	58	121.3	54-198
Distal right	24	110.1*	71-159	12	74.2*	39-247	7	119.2	71-189	58	142.6	76-210
Proximal right	24	85.9	37-143	12	47.3*	33-128	7	73.1	50-148	58	113.1	57-167
<b>Pressure of puborectalis muscle</b>												
Rest	24	52.7*	34-104	12	44.2 *	11-69	7	49.3*	23-79	61	68.9	49-92
Squeeze	24	85.3*	45-145	12	58.9 *	26-137	7	75.5	57-175	58	124.9	70-183

Med – median; pc – percentile; HD – Hirschsprung's disease; AA – anal atresia; PC – proctocolectomy; \* $p < 0.05$  compared to HC group.

**Table IV.** The pressures of segments in patients after surgery compared to the control group.

Segment	HD			AA			PC			HC		
	N	Med	5-95 pc	N	Med	5-95 pc	N	Med	5-95 pc	N	Med	5-95 pc
Mean resting pressure (mmHg)	71	87.5	65-125	20	61.3*	24-116	6	69.9	46-96	7	67.9	51-149
Maximal squeeze pressure (mmHg)	68	207.7	120-292	20	168.1*	60-246	6	153.4	91-338	7	103.8*	62-193
Anal canal length (cm)	71	2.6	1.8-3.8	20	2.3	1.6-2.5	6	2.8	1.5-3.1	7	2.4	1-4
Minimal rectal compliance (cm <sup>3</sup> /mmHg)	69	0.15	0.12-0.27	20	0.16	-27.7-0.3	6	0.17	0.13-0.18	7	0.18	0.12-0.29
Maximal rectal compliance (cm <sup>3</sup> /mmHg)	69	0.62	0.25-2.07	20	0.91	0.29-2.06	6	0.95	0.36-1.94	7	1.2	0.27-1.99
RAIR (cm <sup>3</sup> )	65	10	10-30	8	20	10-30	3	30	10-60	0	-	-
Sensation (cm <sup>3</sup> )	58	20	10-80	13	20	10-60	4	45	10-70	2	35	10-60
Urge (cm <sup>3</sup> )	58	30	10-130	13	35	20-70	4	87.5*	60-179	1	-	-
Discomfort (cm <sup>3</sup> )	58	87.5	20-180	13	60	40-165	4	127.5	60-210	1	-	-

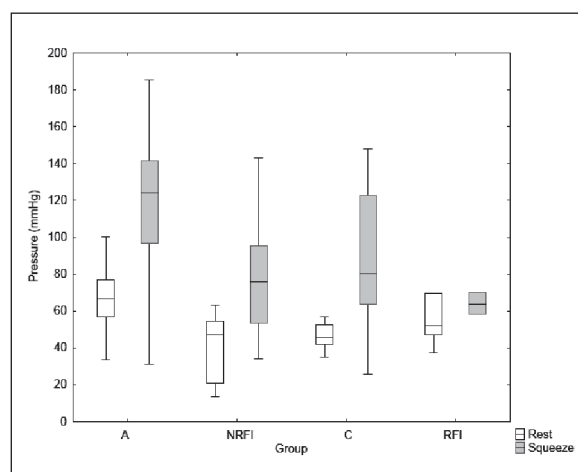
Med – median; pc – percentile; HD – Hirschsprung's disease; AA – anal atresia; PC – proctocolectomy; \* $p < 0.05$  compared to HC group.



**Figure 1.** ROC curve for mean resting pressure and incontinence with cut-off value discriminating between asymptomatic children and patients with fecal incontinence.

might lead to the overlooking of more discrete pressure gaps.

Results of most manometric studies evaluating children after surgery for anorectal malforma-



**Figure 2.** Puborectalis muscle resting and squeeze pressures according to anorectal function (A – asymptomatic; NRFI – non-retentive fecal incontinence; C – constipation; RFI – retentive fecal incontinence).

tions are more consistent. Similar to our results, resting pressures obtained by other authors tend to be lower compared to normative data. This may be the result of the anomalous anatomy of the anal canal. Asymmetry of the anal canal diagnosed by profilometry was often observed in these patients by Vital Júnior et al<sup>33</sup>. They found abnormalities of the pressure profile of more than

**Table V.** Mean pressures of segments in regard to symptoms.

Segment	A			NRFI			C			RFI		
	N	Med	5-95 pc	N	Med	5-95 pc	N	Med	5-95 pc	N	Med	5-95 pc
<b>Rest</b>												
Distal anterior	71	69.4	38-101	20	36.9*	6-73	6	33.7*	0,7-72	5	33.1	15-85
Proximal anterior	71	38.5	15-68	20	29.4	11-65	6	30.9	5-53	5	39.6	25-108
Distal posterior	71	44.3	18-73	20	25.7*	6-64	6	31.5	3-60	5	22.7	-3-85
Proximal posterior	71	68.7	41-98	20	41.7*	13-70	6	42.1*	10-70	5	49.3	32-96
Distal left	71	80.7	54-114	20	50.7*	21-89	6	59.3*	17-85	5	67.8	37-102
Proximal left	71	64.9	35-104	20	45.3*	21-70	6	51.9	13-73	5	63.1	42-109
Distal right	71	81	54-117	20	58.2*	17-94	6	57.3*	20-78	5	67.7	23-120
Proximal right	71	66.6	41-92	20	49.2*	14-78	6	38.5*	19-65	5	50.4	37-106
<b>Squeeze</b>												
Distal anterior	68	118.4	54-195	20	69.6*	29-155	6	82	29-140	5	35.7*	20-94
Proximal anterior	68	61.5	14-119	20	51.5	18-87	6	40.6	11-90	5	29.6	7-163
Distal posterior	68	67.6	25-129	20	57.3	21-109	6	49.8	18-123	5	51.3	14-128
Proximal posterior	68	133.3	48-212	20	82.3*	24-165	6	73.4	43-155	5	66.5*	34-119
Distal left	68	126.5	74-184	20	95.2*	50-170	6	98	47-151	5	74.4	44-135
Proximal left	68	121.1	53-182	20	91.9	34-145	6	79.6	23-155	5	62.1	35-197
Distal right	68	135.6	72-205	20	102.7	54-219	6	90.6	52-159	5	79.1*	39-129
Proximal right	68	112.6	51-164	20	80.8	47-143	6	67.9	37-119	5	58.9*	33-113

Med – median; pc – percentile; RAIR – recto-anal inhibitory reflex; A – asymptomatic; NRFI – non-retentive fecal incontinence; C – constipation; RFI – retentive fecal incontinence; \**p* < 0.05 compared to A group.



half of the investigated patients. This resulted in a high rate of complete incontinence in patients. Caldaro et al<sup>34</sup> investigated the anal canal ultrasonographically and found abnormalities in the morphology of sphincters that were further correlated with a weak resting pressure of the anal canal, which supports the observations in our study.

Little is known about the correlation between function and manometry in pediatric patients after proctocolectomy. In our sample, the mean resting pressure was within a normal range. This may be due to the timing of the follow-up care or the older age of the group compared to other groups. Decreased resting and squeeze pressures may improve during 3 years of follow-up care<sup>35</sup>. Data about the morphological analysis of the anorectum after proctocolectomy in children are lacking. It is known from studies<sup>36-38</sup> in adult patients that the mechanism for continence is addressed to a consistent radial distribution of the pressure of the anal canal.

To produce a stronger statistical analysis of bowel function and manometric data, we reorganized the groups with respect to symptoms. Due to many inconsistent definitions of incontinence and constipation in children after surgery, we decided to use the Rome criteria despite the fact that these criteria pertain to functional disorders without any organic causes. According to the definitions, we divided incontinence into retentive and non-retentive. Therefore, straight comparisons between our results and results from the literature should be taken with caution. We found that patients suffering from fecal incontinence without retention have a significantly decreased resting pressure compared to asymptomatic children. This is consistent with the majority of studies utilizing conventional manometry, in spite of the diversity in the definitions of incontinence. Low resting and squeeze pressures are reported in patients with HD and incontinence<sup>11,39,40</sup>, although not all authors found this difference to be significant<sup>41,42</sup>. More consistent data have been published about incontinence in children after surgery for ARM. Decreased anal pressure was often reported as being correlated with incontinence<sup>43-45</sup> and expected in patients in whom the pressure was below 30% of the reference value<sup>46</sup>. Moreover, decreased pressure during voluntary squeeze was also found which was correlated with continence<sup>47,48</sup>.

A possible mechanism of incontinence in patients after surgery suggested in the literature

is a shorter anal canal length<sup>49</sup>. In our study, we did not find any difference in the length of the anal canal between the patients and the healthy controls. Parameters that were correlated with incontinence were addressed only to the pressure values.

Notably, we observed a high rate of incontinence in patients after proctocolectomy. Contrary to our results, a good functional outcome is reported in the literature<sup>35,50</sup>. The difference may be the result of different surgical procedures. In our study, all of the patients had ileoanal anastomosis, while in other studies, ileal-J-pouch anastomosis was preferred. Pouches are considered to be good reservoirs for feces, preserving the continence mechanism. Rink et al<sup>12</sup> in adults comparing these two anastomotic techniques have shown that good continence is present not only in patients with proper resting pressure but also in those with adequate rectal compliance and greater values of maximal tolerated volume observed, which was more likely observed in patients with pouches.

In our study, children suffering from retentive fecal incontinence had decreased pressure parameters (conventional and segmental) only in the squeezed state. This suggests the possible role of striated muscles, the puborectalis muscle (PRM), and the external anal sphincter (EAS). The role of EAS in incontinence was recently suggested for women after obstetric trauma. The authors<sup>51</sup> stated that its purse-string morphology may play a crucial role in partial continence. In women with only unilateral EAS damage, the whole muscle may be compromised.

As seen in our study, thresholds of sensation are correlated with the continence mechanism. In constipated patients, we observed higher thresholds. This may be the result of rectal distension. Data from studies<sup>36,52,53</sup> in adults suggested that the number of stools per day is correlated with the thresholds of sensation and rectal compliance.

Lower levels of thresholds observed in incontinent patients may be the result of the shorter length of the bowel left after surgery<sup>54-60</sup>. Another possible mechanism has been suggested in adult studies. Fruehauf et al<sup>61</sup> showed the interaction between the anal pressure and the thresholds of sensations in healthy adults. Decreased thresholds were observed in individuals in whom lower resting pressures were recorded. This phenomenon may be a reaction to the lower pressure of damaged anal sphincters and may represent a

form of compensation mechanism. The authors<sup>61</sup> suggest that damage to this mechanism may result in fecal incontinence. In our study, this mechanism may work improperly because of the anomalous nervous system, which may be due to the malformation of the anorectum. Moreover, as it has been shown in adults<sup>62,63</sup>, the function of sacral and pudendal motor nerves may be abnormal in incontinent patients after surgery. This may explain the lack of statistical significance of thresholds in the incontinent patients in our group, although an overall tendency toward diminished values was found.

A poor functional outcome after surgery may be the result of improper functioning of the colon, as was reported in children after surgery for HD<sup>64,65</sup>, or the presence of dyssynergic defecation in children after surgery<sup>66</sup>. In children with the proper function of the anal sphincters, other mechanisms should be considered, and further tests are advised<sup>8,9</sup>.

The major advantage of our study is that we evaluated children after surgical procedures using precise technology that proved to be safe, well-tolerated, and useful in a group of patients with protracted symptoms after surgery. The extent of disturbed sphincter function suggests that tests of anal structure (ultrasonography/magnetic resonance imaging) should always be considered in patients with protracted symptoms of incontinence, especially if surgical intervention is being considered. Otherwise, other functional tests should be taken into account, such as colonic transit or tests of sensation (rectal barostat).

Our research has some limitations. The study sample was heterogeneous, and therefore, the subgroups were relatively small. Additionally, the PC group was older than other groups, which could have an influence on the direct comparison of pressures between groups after surgery but not on comparisons with the HC group. Moreover, the surgical techniques differed among patients, making it impossible to find characteristic patterns in segmental pressure gaps correlated with a particular surgical method. Despite this, we were able to show the vast extent of the disturbances in pressure profiles in these patients. The significant differences between subgroups in the conventional and segmental pressure parameters highlight the key role of anal sphincters and rectal sensation in the generation of symptoms. Another limitation is that we did not perform other tests that can identify additional factors contributing to poor continence, such as colonic

transit studies or colonic manometry. Still, the extent of the sphincter dysfunction in our sample underlines the importance of the anorectum in the generation of symptoms and emphasizes that the test of the anal function should be performed first<sup>8</sup> and addressed as a first-line treatment. Another limitation is that the comparison of the pressure topography of the anal canal after surgery for ARM may be controversial due to possible different anatomies of the neorectum in particular patients. The pressure of PRM may be estimated with some degree of error, especially in patients with a high type of anorectal malformation. Despite that, we found an overall decreased pressure of the anal canal, which was correlated with incontinence. The pressure of PRM should be discussed with caution, and the location of this muscle should be established by ultrasonography. Another limitation is that we did not analyze the time of follow-up after surgery, which might have affected the pressure readings. The last limitation of this study is the size of the probe. A greater catheter diameter in relation to a smaller size of the anal canal may produce higher pressure readings in younger children.

## Conclusions

Our study demonstrated lower pressure parameters in children after surgery, with the lowest values found in patients suffering from anal atresia. Decreased conventional and segmental pressure parameters are responsible for fecal incontinence. We showed that 3DHRAM may be a safe and useful tool for assessing the function of the anorectum of children after surgery.

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### Conflict of Interest

The Authors declare that they have no conflict of interests.

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### Authors' Declaration of Personal Interests

Marcin Banasiuk has received equipment support for study and speaker honorarium from Medtronic; Aleksandra Banaszekiewicz, Marcin Dziekiewicz, Łukasz Dembiński, Dariusz Piotrowski, Andrzej Kamiński have no conflict of interest.

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The Authors declare that they have no conflict of interests. Medtronic (Ireland) provided manometric equipment (catheter and disposable sheaths).

**Authors' Contribution**

All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Marcin Banasiuk, Marcin Dziekiewicz, Łukasz Dembiński, Dariusz Piotrowski, Andrzej Kamiński and Aleksandra Banaszkiwicz. The first draft of the manuscript was written by Marcin Banasiuk and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

**Ethics Approval**

This study was approved by the Ethics Committee of the Medical University of Warsaw, Poland (KB7/2013). The procedures used in this study adhere to the tenets of the Declaration of Helsinki.

**Clinical Trials Registration**

The study was registered at ClinicalTrials.gov.

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