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*Original Article*

# Application of Three Dimensional High Resolution Anorectal Manometry to Demonstrate Anal Sphincter Pressure Asymmetry in Children after Anorectal Surgery: A Pilot Study

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**Abstract** ***Purpose***: The use of anorectal manometry in the evaluation of anal sphincter function has been well established. The new three dimensional (3D) high resolution anorectal manometry (HRARM) would provide additional information regarding the symmetry of the anal sphincter pressure. ***Methods***: We prospectively recruited 17 patients with defaecation disorders: 3 patients with idiopathic constipation, 8 patients with Hirschsprung's disease and 6 patients with anorectal malformation. 3D HRARM was performed. The resting pressure and squeeze pressure at the physiological sphincter were measured and analyzed. We define the inter-quadrant pressure asymmetry index (Δp) mathematically, Δp = (maximal pressure - minimal pressure)/maximal pressure x 100%. ***Results***: There was an increasing inter-quadrant pressure asymmetry index (Δp) observed in the anal pressure profiles in patients with history of Soave pullthrough, myomectomy, Duhamel operation and posterior sagittal anoplasty (PSARP). The asymmetry is more pronounced in squeeze pressure in the Soave pullthrough group, and less pronounced in squeeze pressure in the myomectomy, Duhamel and PSARP group. ***Conclusions***: The inter-quadrant pressure asymmetry index (Δp) reflects the post operative status of anal sphincter, which is influenced by the different surgical maneuvers employed in the operations. The symmetry of anal sphincter pressure might provide useful anatomical parameters in the assessment of surgical outcomes.

**Key words** Anorectal malformation; Anorectal manometry; High resolution; Hirschsprung's disease; Symmetry

## Introduction

Anorectal manometry (ARM) has been used extensively in research and in the clinical assessment of patients with defaecation disorders. It has been shown that it can provide reliable analysis of the anal sphincter function.[1,2] There are increasing reports of ARM application on paediatric population.[3,4]

In conventional ARM, a trans-anal water-perfused multi-channel catheter is inserted into the rectum. Serial measurements of resting, squeeze and push pressures at different levels of anorectal region are measured. The catheter is then manually withdrawn by a pull-through technique until it reaches the anal sphincter region.

However, the frequent manipulation of the catheter may be disturbing to a young child, leading to inaccurate results and conclusions. With the evolution of technology, the three dimensional high resolution anorectal manometry (3D HRARM) has been released to the market. It has radially arranged pressure channels in multiple levels; hence the anal pressures along the entire anal canal can be measured simultaneously, obliterating the need of frequent adjustment of the catheter during the procedure. The individual pressure parameters can be integrated by the computer software to

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reconstruct a topographic image of anal canal.

The aim of this study is to investigate the feasibility and usefulness of 3D HRARM in the evaluation of patients who had undergone surgery for congenital anorectal anomalies including Hirschsprung's disease (HD) and anorectal malformation (Mal).

## Materials and Methods

From June 2013, we prospectively recruited 14 neurologically healthy children with age 4 years or above, with surgical correction of HD and Mal in our hospital.

All the patients were recruited at our multidisciplinary bowel management clinic. Their demographic data and Rintala continence scores are shown in Table 1.[5] Patients with history of HD were further analysed into subgroups according to the surgical techniques employed: five patients had laparoscopic assisted Soave endorectal pullthrough for rectosigmoid HD, two patients had posterior myomectomy for ultra-short segment HD and one patient had Duhamel operation for long segment HD. All the patients in the Mal group had anorectal malformation with posterior sagittal anorectoplasty (PSARP) performed.[6] Three patients with idiopathic constipation were recruited as control group.

With informed consent, all the recruited subjects were given bowel preparation in the form of sodium phosphate rectal fleet enema 3 ml/kg prior to the study. The caretaker was allowed to accompany the child during the procedure to relieve anxiety. A surgeon was present during the entire procedure to conduct and analyse the pressure tracings. A single-use water perfused 24-channel catheter (Figure 1) with four quadrant pressure channels at five 1 cm spacing (Medical Measurement Systems B.V., MMS G-90520 The Netherlands) was used for manometry.

All the patients were awake and non-sedated during the investigation. The physiological anal sphincter was identified at the high pressure zone (HPZ) of the anal canal. The catheter would then be held in place by securing it onto the subject's buttock with surgical tape. The resting and squeeze pressure at the HPZ were measured.

The orientation and position of the catheter is checked regularly after each manoeuver to avoid rotation. The pressure parameters were integrated into a topographic image of the anal canal by the 3D HRARM software.

PASW Statistics 18 (SPSS Inc, Chicago, IL, USA) was applied for statistical analysis. Wilcoxon signed ranks test was used to compare non-parametric data, with statistical significance considered at $p<0.05$. The study was approved by the research ethical committee of the hospital.

## Results

All the patients enrolled were able to complete the manometric study with no complications. There is no up-to-date validated data concerning the symmetry of the pressure distribution of the anal canal in children using 3D HRARM. In our study, we define the inter-quadrant pressure asymmetry index (Δp). Δp denotes the inter-quadrant pressure difference divided by the maximal quadrant pressure, in percentage expression.

Δp = (maximal quadrant pressure - minimal quadrant pressure)/maximal quadrant pressure x 100%.

For total symmetrical sphincter with equal pressure over the four quadrants, Δp = 0%. For total asymmetrical sphincter, Δp = 100%.

In the control group, Δp ranged from 8-23% in the resting pressure (Figure 2). In the HD group, patients with laparoscopic assisted Soave pullthrough had Δp range from 27-50%; patients with myomectomy and Duhamel operation had Δp range from 56-95%. In the Mal group, patients had Δp range from 59-95% (Figure 3). However, we do not find any correlation of the inter-quadrant pressure asymmetry index with the Rintala score (Table 2).

The squeeze pressures at the HPZ were also measured

**Table 1** The demographic data and Rintala continence scores of patients

| Study groups | Control | Hirschsprung's disease | Anorectal malformation |
|---|---|---|---|
| Gender | 3M: 0F | 4M: 4F | 2M: 4F |
| Age (years) | 14 (10-17) | 8 (6-10) | 5(4-9) |
| Rintala score (/20) | 16 | 17.6 | 18.3 |

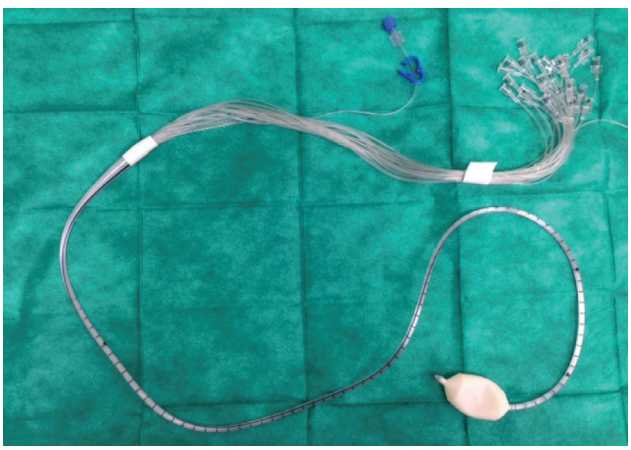

**Figure 1** Three-dimensional high resolution anorectal manometry catheter.

and the inter-quadrant squeeze pressure asymmetry index (Δp') was expressed as the inter-quadrant pressure difference divided by the maximal quadrant pressure on squeezing. Δp' were significantly greater than Δp in patients with idiopathic constipation or Hirschsprung's disease with laparoscopic assisted endorectal pullthrough performed (Wilcoxon signed ranks test, p=0.049). However, Δp' were significantly smaller than Δp in patients in HD group after myomectomy/Duhamel operations or Mal group after PSARP (p=0.021).

## Discussion

HD and Mal are two of the most common congenital anorectal anomalies. Defaecation disorders such as faecal incontinence and constipation can occur after the surgical repair of these conditions. Most of these patients require long-term bowel management program. An objective evaluation of their sphincter function is essential in the evaluation of surgical outcomes. ***In this study, we limit our application of the manometric study to patient older than 4 years old, because when ARM study is applied in children younger than 4 years old, ketamine sedation is often required because they are too young to cooperate with the study. In the ideal situation, the control group should be children with no gastro-intestinal symptoms (no constipation), however it would be difficult to recruits such children as they would not even come to see us in the first place.***

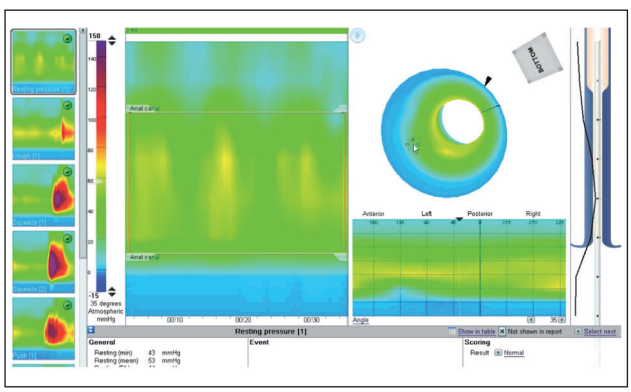

**Figure 2** Topographic image of anal canal in patient with idiopathic constipation (rest).

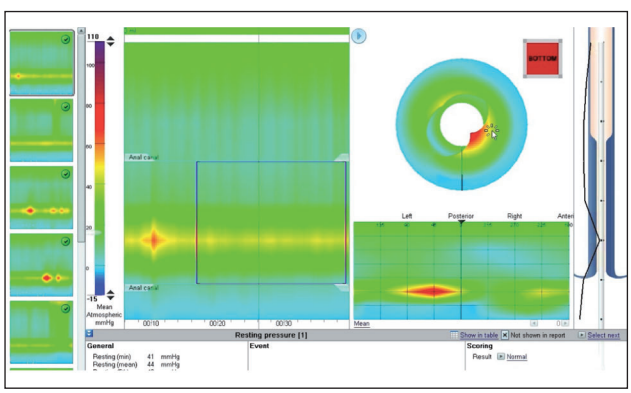

**Figure 3** Topographic image of anal canal in patient with anorectal malformation after posterior sagittal anorectoplasty (rest).

Faecal continence is significantly correlated with anal pressures in post-operative patients.[7,8] Various studies have also discussed the functional outcome of post-operative patients by means of validated questionnaires and/or bowel function scoring system. Few reports have evaluated the anatomy of the anal canal with endorectal ultrasound and MRI pelvis.[9] Sphincter asymmetry index (SAI) have been described and used for the detection of sphincter defect[10,11] and to assess the integrity of sphincter after operation for HD,[12] however these studies are often based on adults while using the conventional anorectal manometry.

Recently, the availability of the 3D HRARM has allowed a detailed assessment of pressure distributions in the anal canal. Cheeney et al[13] have suggested that 3D HRARM could improve the understanding of anorectal physiology by permitting concurrent assessment and correlation of anatomy with anal pressures profiles, eliminating the need for subsequent correlation with other imaging modalities. Nonetheless, the experience of using 3D HRARM in the paediatric surgical patients is limited. To the best of our knowledge, this is the first study to evaluate the anal pressure in post-operative paediatric patients by 3D HRARM.

To facilitate communication and understanding, we derived a simplified mathematical model by calculating the pressure difference between the quadrant with the maximal pressure and the quadrant with the minimal pressure, and then we quantify the inter-quadrant pressure asymmetry index (Δp) by taking the pressure difference in relation to the maximal pressure, the higher value of Δp, the more the asymmetrical is the anal canal.

We observed that there is a trend of mild asymmetry in the anal pressures in the control group, with Δp <25%; moderate asymmetry in the Soave HD group with Δp ranges from 25%-50%; and marked asymmetry in the Duhamel HD, myomectomy HD and Mal group with Δp >50%.

Ambartsumyan et al[14] had defined the symmetry of the anal canal by incorporating the longitudinal and radial anal pressure distribution, which necessitate the measurement and adjustment of the anal canal length. In our investigations,

we did not take into account of the length of the anal canal as we only focused on the degree of asymmetry of the high pressure zone (HPZ). As we believe that HPZ is the most representative for sphincter function. The variable body size in the paediatric population[15] and the different surgical procedure that had been performed may also lead to imprecise and difficult interpretation of the genuine canal length.

Several studies had discussed the significance of anal pressure asymmetry in nulliparous adult women and it was suggested that the asymmetry was due to the unique "triple looped anatomy" of the puborectalis muscles in the anal canal, which was evident in both high resolution anal manometry and endorectal ultrasound.[16,17] Hence, the anal pressure profiles of the control group in our study are also not completely symmetrical.

In Soave pullthrough operation, submucosal dissection of the anal cuff with the preservation of anal sphincter was performed. However, we believe inadvertent microscopic damage of the circular fibers may lead to moderate asymmetry in anal pressures.

In both myomectomy and Duhamel operation, the posterior part of the anal sphincter was disrupted, that may account for the moderate to marked asymmetry in anal pressures.

In the Mal group, we expected that by following Pena's principle of adhering to the midline during dissection and by accurate siting of the neo-anus with the nerve stimulator when performing PSARP would preserve the anal sphincter muscle complex. Unfortunately, marked anal pressure asymmetry was found in this group of patient, we postulated that it could be due to the congenital defects of the neuromuscular complex in the anal canal or muscle injury during operation.

It is interesting to note that in constipation and Hirschsprung's disease patients after laparoscopic assisted Soave endorectal pullthrough with mild to moderate anal sphincter asymmetry, Δp' was significant greater than Δp. On squeezing of anal sphincter, contraction of the unique "triple looped puborectalis" muscles in the anal canal may account for the exacerbation of the sphincter asymmetry. However, in patients suffering from Hirschsprung's disease

**Table 2** The results of the anal sphincter pressure recorded by the 3D HRARM

| Subject | Diagnosis (Operation) | Resting pressure (mmHg) (max./min.) | Degree of asymmetry (%) (resting pressure)(Δp) | Squeeze pressure (mmHg) (max./min.) | Degree of asymmetry (%) (squeeze pressure) (Δp') | Rintala score |
|---|---|---|---|---|---|---|
| 1 | Constipation | 79/67 | 15 | 215/161 | 25 | 13 |
| 2 | Constipation | 70/54 | 23 | 297/151 | 49 | 19 |
| 3 | Constipation | 74/60 | 8 | 134/113 | 15 | 18 |
| 4 | HD (Soave) | 70/43 | 39 | 158/56 | 64 | 15 |
| 5 | HD (Soave) | 70/43 | 39 | 302/147 | 51 | 18 |
| 6 | HD (Soave) | 79/58 | 27 | 126/74 | 41 | 17 |
| 7 | HD (Soave) | 53/72 | 49 | 198/123 | 38 | 19 |
| 8 | HD (Soave) | 109/55 | 50 | 116/56 | 52 | 18 |
| 9 | HD (myomectomy) | 85/11 | 87 | 103/20 | 81 | 18 |
| 10 | HD (myomectomy) | 81/36 | 56 | 206/163 | 21 | 18 |
| 11 | HD (Duhamel) | 113/3 | 95 | 137/35 | 75 | 19 |
| 12 | Mal (PSARP) | 86/5 | 94 | 124/68 | 45 | 20 |
| 13 | Mal (PSARP) | 85/11 | 87 | 103/20 | 81 | 18 |
| 14 | Mal (PSARP) | 70/25 | 64 | 181/63 | 65 | 17 |
| 15 | Mal (PSARP) | 113/6 | 95 | 137/35 | 75 | 17 |
| 16 | Mal (PSARP) | 92/21 | 78 | 73/14 | 81 | 18 |
| 17 | Mal (PSARP) | 73/30 | 59 | 238/121 | 49 | 20 |

HD: Hirschsprung's disease; Mal: anorectal malformation; PSARP: posterior sagittal anorectoplasty

after myomectomy / Duhamel operation and anorectal malformation after PSARP with marked sphincter asymmetry, Δp' was significant smaller than Δp. The augmenting effect of pelvic floor and gluteal muscles contraction during anal squeeze may account for the compensation of weaken quadrant of anal sphincter.

Based on the above findings, we conclude that the degree of asymmetry (Δp) can reflect the post-operative status of anal sphincter, which is largely influenced by the specific surgical maneuvers employed in the operations for HD and Mal. Therefore, we infer that the study of the pressure distribution in the anal canal can provide useful feedback information to the surgeons and might facilitate subsequent refinement of the surgical techniques.

In this pilot study we were not able to find a clinical correlation of the sphincter pressure status and the functional outcome (in terms of Rintala scores) of these patients. This may also be due to the different response and compliance to the post op bowel management program[18,19] administered to all our patients after anorectal operations.

A significant limitation of our study lies in the small sample size, with increasing experience and expertise in the application of 3D HRARM, we hope that we can eventually establish some normative data in the assessment of anal sphincter physiology.

The use of 3D HRARM in the assessment of postoperative anal sphincter condition is safe and child-friendly. It may give additional information regarding the degree of the anal pressure symmetry. With further studies and correlation with other investigation modalities, it would become an increasingly useful armamentarium of investigations in the assessment of children with defaecation disorders.

## Declaration of Interest

None

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