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Size of Functional Sphincter Defect after Repair of Anorectal Malformations Measured by Three-Dimensional High Resolution Anorectal Manometry Correlates to Fecal Incontinence

Meyer TB[1,2], Christensen P[3], Jakobsen M[4], Baatrup G[1,2] and Qvist N[1,2]*

[1]Department of Surgery, Odense University Hospital, Denmark

[2]Department of Clinical Research, University of Southern Denmark, Denmark

[3]Department of Surgery, Arhus University Hospital, Denmark

[4]Department of Pediatrics, Odense University Hospital, Denmark

Abstract

Purpose: Anorectal Malformations (ARM) are a spectrum of anomalies of the rectum and anal canal affecting 1 in 2500 to 5000 live births. Functional problems are common and related to the type of ARM and associated malformations. We aimed to evaluate the results of Three-dimensional High Resolution Anorectal Manometry (3D-HRAM) in long-term follow-up after surgical correction of ARM with special reference to fecal incontinence.

Methods: Twenty-one patients with anorectal malformations and primary repair at our center consented to participate in the study. Pressures of the anal sphincter muscles and defects were addressed by 3D-HRAM. Fecal incontinence and disease-specific quality of life were evaluated by the fecal incontinence quality of life score and Wexner incontinence score respectively. The study was approved by the Committee in Health Research Ethics and the Danish Data Protection Agency (S-20140017).

Results: Median age was 22 (12 to 31) years and 13 (67%) participants were females. Sphincter defect was present in 48% (N=10) of participants. Participants with sphincter defects had significant higher Wexner score and size of sphincter defects was correlated to Wexner score. Participants with or without sphincter defects did not differ on manometry parameters including resting anal and squeeze pressure or disease-specific quality of life.

Conclusion: In a study of the long-term outcome after repair of anorectal malformations we found a higher Wexner incontinence score in the presence of an anal sphincter defect and the size of the defect was correlated to the Wexner incontinence score.

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Correspondence:

Meyer TB, Department of Surgery, Odense University Hospital, Odense C, 5000, Denmark, Tel: +45 27896080; E-mail: thomas.bjoersum-meyer@rsyd.dk

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Introduction

Anorectal Malformations (ARM) are a spectrum of anomalies of the rectum and anal canal affecting 1 in 2500 to 5000 live births [1,2]. Even after proper reconstruction, functional problems are common and related to the type of ARM and associated malformations. Only few studies have addressed the findings on advanced anorectal manometry in relation to the long-term functional outcome [3]. Conventional anorectal manometry has gained widely use and acceptance for the evaluation of the anorectal function in different types of anorectal malformations [4]. The results have been contradictory and with dubious clinical value. Some studies have found a correlation between the extent of sphincter damage and the degree of continence and others have not [5]. Three-Dimensional High Resolution anorectal manometry (3D-HRAM) is a technique providing an image of the pressure profile of the sphincter complex. In patients with fecal incontinence of other causes, 3D-HRAM has shown a high negative predictive value for the finding of an acquired anatomic sphincter defect compared to EUS [6]. To our best knowledge no previous studies has addressed the value of the 3D-HRAM anal manometry in the long-term follow-up after surgery for ARM.

The aim of the present study was to evaluate the results of 3D-HRAM in the long-term follow-

up after surgical correction of ARM with special reference to the status of anal continence.

Materials and Methods

Subjects

Participants were recruited from a tertiary pediatric surgical center at Odense University Hospital (OUH), Denmark with a catchment area of 3.5 million people for the treatment of ARM. Subjects were identified through local databases using the ICD-9 codes: 725.1, 725.2 (1985-1994) and ICD-10 codes: Q42 (all included) and Q438K (1995-2004). Inclusion criteria were primary surgery for anorectal malformations from 1985 to 2004 at OUH. Exclusion criteria were cognitive disability to an extent that the patient could not properly understand purpose and consequences of the study. An invitational letter was sent to each subject and if no response was registered, another letter was sent 14 days later. A total of 21 subjects fulfilled inclusion criteria and consented to participate.

Data obtained from patient charts were: age, comorbidity, and type of malformation, type of primary surgery for anorectal malformation, other known congenital abnormalities or malformations and, in women, childbirth.

The study was conducted according to the 7th revision of the Helsinki declaration (2013). Verbal and written informed consent was obtained from adult participants and in participants below 18 years of age, from parent(s) or guardian(s). The study was approved by the National Committee on Health Research Ethics and the Danish Data Protection Agency (S-20140017).

High resolution anorectal manometry

We used a Manoscan™ anorectal High Resolution Manometry system (Medtronic, MN, USA) mounted with a 3D probe. The rigid probe had 256 pressure sensors circumferentially aligned over the length of 64 mm and a circumference of 10 mm. The probe was calibrated before use in each participant. A disposable sheath with a rectal balloon was applied and lubricated before introducing the probe into the anus. Neither enema or colonic preparation nor sedatives were used before examination. Participants were placed in the left decubitus position with hips and knees flexed 90 degrees during the procedure. Before introducing the probe, a rectal examination was done to ensure the rectum was empty. The same clinician (TBM) with experience in performing anorectal manometries performed all examinations.

After introducing the probe, a resting period of one minute was awaited before measurements. The resting pressure was measured continuously during a period for 20 sec three times. The mean value was used for the calculation. Squeeze pressure and push maneuvers were then performed three times with 30 sec intervals and mean value was used for the calculation. In order to elicit the recto-anal inhibitory reflex the rectal balloon was forcefully inflated with increasing volumes of air in 10 ml aliquots stooping at 60 ml if the reflex was not elicited. Rectal sensitivity was examined by consecutively inflating aliquots of 10 ml of air until first sensation, urge to defecate and discomfort was registered. The procedure was stopped at a volume of 400 ml. The high-pressure zone area was calculated as (rectal pressure + (anal resting pressure – rectal pressure)) × 0.25). The presence, size and location of a sphincter defect were calculated from the 3D presentation of the anal resting pressure profile (Figure 1). A functional anal sphincter defect was defined as a pressure below 25 mmHg and the size of the defect was calculated by multiplying the percentage of the anal circumference with a pressure measurement below 25 mmHg by 360°.

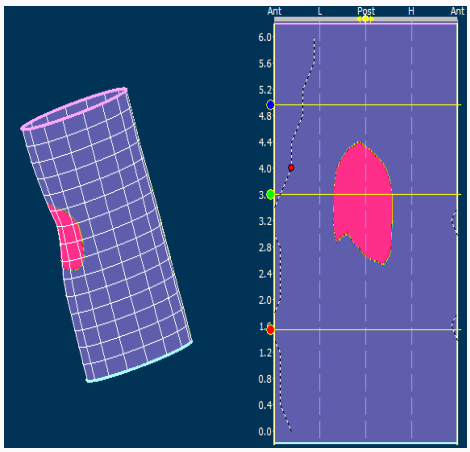

Figure 1: Examples of anal resting pressure profile in two participants. The 3D presentation is on the left and the 2D presentation is on the right. Pink color represents pressures equal to or above 25 mmHg. A: No complete sphincter defect is present. B: Large complete sphincter defect is found and only a part of the posterior sphincter complex is intact. Numbers on the left margin of the 2D presentations indicate length in centimeters. Ant: anterior. L: left. Post: posterior. H: right aspect of the sphincter complex.

Data and statistics

Data from High Resolution Anorectal Manometry were analyzed using the guide function in the Manoview® 3.0 software. We studied the following variables: mean resting pressure, mean squeeze and maximum squeeze pressures, mean rectoanal pressure difference during push maneuver, high anal pressure zone, air volumes for eliciting recto-anal inhibitory reflex and air volumes for first sensation, urge to defecate and discomfort. A non-normal distribution of data was assumed. Data were presented as medians and Interquartile Ranges (IQR) if not otherwise indicated. Discrete data were compared with Fisher`s exact test and for continuous data the Mann-Whitney U test was applied. Level of significance was reported with two-sided p-value. Correlation tests were performed with the Spearmann`s rank order correlation. Positive correlation was reported with a correlation coefficient rho (ρ).

Questionnaires

Soiling and constipation were evaluated with the Krickenbeck Classification [7] of postoperative results (Table 1). Wexner score was used to address severity of fecal incontinence [8]. Disease - specific quality of life was evaluated by the Fecal Incontinence Quality of Life (FIQL) -score. The FIQL -score is based on four scales; lifestyle, coping/behavior, depression/self perception and embarrassment [9].

Subject characteristics

Table 1 shows subject characteristics. In two participants we were unable to perform HRAM because of anal stenosis and they were omitted from this study. Of the 21 included participants five were below 18 years of age. None of the female participants have had a vaginal delivery. The anocutaneous fistula was the most common type of anomaly. In 43% (9/21) of subjects, associated anomalies appeared. Four subjects had more than one associated anomaly. Low malformations were anocutaneous fistula (N=8), anal stenosis (N=1) and anovestibular fistula (N=1). Group with intermediate

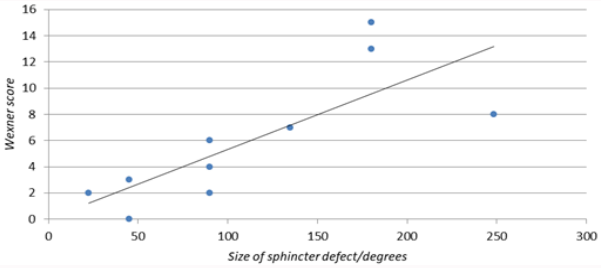

Figure 2: Scatterplot of Wexner *vs*. circumferential size of sphincter defect.

Table 1: Krickenbeck classification of postoperative results.

Voluntary bowel movements	Yes/no
Feeling of urge, capacity to verbalize and able to hold bowel movements.	
Soiling	Yes/no
Grade 1: Occasionally (once or twice per week)	
Grade 2: Every day, no social problem	
Grade 3: Constant, social problem	
Constipation	Yes/no
Grade 1: Manageable by changes in diet	
Grade 2: Requires laxatives	
Grade 3: Resistant to laxatives and diet	

malformations consisted of vaginal fistula (N=4), rectobulbar fistula (N=4) and rectovestibular fistula (N=1). Two participants had high malformations, one with no fistula (N=1) and one with a cloaca malformation (N=1).

Results

Functional results

The incidence and severity of soiling and constipation are presented in Table 2. Both soiling and constipation were present in approximately half of the participants. Only one patient experienced soiling as a social problem (grade 3). None of the participants reported constipation resistant to treatment with diet and laxatives (grade 3).

Manometry

In approximately half of the subjects (N=10) a sphincter defect was detected by HRAM and in 30% the defect affected more than half of the sphincter circumference. Largest sphincter defect amounted 249 degrees and the smallest 23 degrees. Median circumferential sphincter defect was 90°. Subjects with lower ARM had significant higher anal resting pressure and anal squeeze pressure compared to subjects with intermediate ARM.

One participant was unable to distinguish between first sensations, urge and discomfort in rectal sensitivity test and in one patient first sensation and urge appeared at the same volume.

Participants with a sphincter defect, was similar to participants without sphincter defect regarding age, gender, resting pressure, squeeze pressure, rectoanal pressure gradient and HPZ as presented in Table 4. Disease-specific quality of life (FIQL) -score did not differ by any scale but Wexner score was significant higher in participants with sphincter defect.

In participants with sphincter defect, more suffered from soiling compared to participants without sphincter defect (P=0.009).

Table 2: Subject characteristics.

Age in years, median and range	22 (12-31)
Female, n and (%)	14 (67)
Appendicostomy, n and (%)	3 (14)
Type of anorectal malformation, n and (%)	
Anocutaneous fistula	8 (38)
Rectourethral fistula (bulbar)	4 (19)
Rectovestibular fistula	2 (10)
Rectovaginal	4 (19)
Anal stenosis	1 (5)
No fistula	1 (5)
Cloaca	1 (5)
Associated anomalies, n and (%)	8 (38)
Vaginal septum	1
Portio duplex	1
Ventricular septal defect	1
Persistent ductus arteriosus	1
Renal agenesis	1
Hydronephrosis	1
Microencephalia	1
Clubfoot	1
Syndromes, n and (%)	2 (10)
Trisonomi 22	1
Caudal regression syndrome	1
Primary surgical approach, n	
Posterior Sagittal Anorectalplasty (PSARP)	11
Perineal reconstruction	5
Dilatation	3
Abdominoperineal pull-through	1
Cutback	1

Table 3: Incidence of bowel symptoms according to the Krickenbeck classification.

Bowel symptom	n (%)
Voluntary bowel movements	7 (33)
Soiling	9 (43)
Grade 1	7 (33)
Grade 2	1 (5)
Grade 3	1 (5)
Constipation	11 (52)
Grade 1	7 (33)
Grade 2	4 (19)
Grade 3	-
Soiling + Constipation	6 (28)

Participants with low malformations had lower anal resting pressure and squeeze pressure ($p<0.05$). No differences were found regarding age, gender, and anorectal pressure gradient, size of HPZ, appearance or size of functional sphincter defects between participants with low and intermediate malformations. One subject without a fistula and one patient with a cloaca malformation were not included in this calculation.

Table 4: Comparison of subject with anal sphincter defect and subject without anal sphincter defect detected by High Resolution Anorectal Manometry.

Parameter	Sphincter defect	No sphincter defect	P-value
Age (years)	23 (12-31)	22 (17-31)	NS
Gender (% female)	60	72	NS
Resting anal pressure (mmHg)	35 (32-90)	37 (33 76)	NS
Anal squeeze pressure (mmHg)	92 (16-251)	178 (56-227)	NS
Rectoanal pressure gradient (mmHg)	-16 (-73-88)	13 (-17-41)	NS
HPZ (cm)	2.6 (1.3-5.5)	3.4 (2.6-4.6)	NS
FIQL score*			
Lifestyle	3.9 (2.2-4)	3.9 (3.3-4.0)	NS
Coping/Behavior	3.4 (1.2-4)	3.7 (3.1-4.0)	NS
Depression/Self perception	3.3 (1.9-4.3)	3.3 (4.4-2.1)	NS
Embarrassment	3.5 (2.7-4)	3.3 (4-1.6)	NS
Wexner score	8 (6-14)	2 (0-7)	0.03

Data are presented as medians (interquartile range) if not otherwise indicated. HPZ: High Pressure Zone; NS: Not Statistical Significant. *FIQL: Fecal Incontinence Quality of Life.

Figure 2 presents a scatter plot of Wexner score *vs*. circumferential size of sphincter defect. A positive and statistical significant correlation was found between Wexner score and size of sphincter defect (ρ 0.87, P 0.001). No other correlations between the results from the manometry and the Wexner score were found.

Discussion

In our study approximately half of the participants (48%) had an anal sphincter defects detected by 3D-HRAM after repair of anorectal malformations. We found the presence and size of functional sphincter defects correlated to the Wexner incontinence score. Endosonography has been the gold standard for assessing the anatomical integrity of the anal sphincter after surgery for anorectal malformations [10]. The problem is that, although the structural defects may be important in the consideration of sphincter repair, the functional defects may explain the relative poor outcome in re-do procedures [11]. In a study by Caldaro et al. [12], internal anal sphincter lesions were found to be common after surgical repair of anorectal malformations. It was detected in 60% of patients with low and intermediate malformations using 3D-endoanal ultrasound (3D-EAUS). The study included 17 children after surgical repair with posterior sagittal anorectoplasty. Six out of 10 patients with low or intermediate malformations had a sphincter lesion, which they deemed to be confined to Internal Anal Sphincter (IAS). However, an anatomic distinction between the different elements of the sphincter complex after PSARP may be difficult and the clinical importance of this is unknown especially for the minor defects. The 3D-HRAM examination gives detailed information on the function of the sphincter complex, which was correlated to functional outcome in our study. The correlation between anal defects shown by 3D-HRAM and defects by ultrasonography or MRI in patients operated for ARM has yet to be proven. We were not able to make an anatomic distinction between the different elements of the sphincter complex with 3D-HRAM but nonetheless able to correlate sphincter defects to functional outcome.

In former studies with conventional manometry a correlation between manometric parameters and clinical outcome has been found. Emblem and colleagues assessed postoperative anatomy in 40 patients with 2D-EAUS after repair of anorectal malformations and compared it to 20 healthy participants [10]. The mean age at follow-up was 16 years (range 1 to 22). Gender distribution was not described. Twenty-five of the 40 included patients were characterized with a high malformation. In our study only two participants were registered with high malformations. An average defect of 2.5 and 3 quadrants in the internal (IAS) and the External Sphincter (EAS) was found respectively. A four level continence scoring system was applied ranging from one to four. Grade four incontinence was defined as incontinence for gas, loose and solid stool. The extent of muscle defect in the IAS was found to be correlated with the degree of incontinence.

In a study by Rezaie et al. [6] 39 adult patients with fecal incontinence but with no previous history of ARM underwent EAUS and 3D-HRAM. Mean age was 65 years. Thirty-one patients were females (79%) and approximately two out of three had had at least one vaginal delivery. Defects were defined as any pressure measurements below 25 mmHg equal to pressure limit for detecting anal sphincter defect in our study. The negative predictive value of HRAM in detecting sphincter defects was found to be 92%. Unfortunately there is no information on any correlation between findings from HRAM and functional outcome. In a similar study where the cut-off value for sphincter defects was a circumferential area with pressure below 10 mmHg the sensitivity and specificity of detecting EAS defects by 3D-HRAM were 65%. In the study by Vitton et al. [13] no such correlation was found. It is generally accepted that the resting anal pressure is maintained by the IAS accounting for about 55% with the EAS and hemorrhoidal cushions adding 30% and 15%, respectively [14,15]. The anatomy and function of the pelvic flour play an important role for the size of squeeze pressure [16]. Thus, a comparison of the results of HRAM between ARM patients with others should be interpreted with caution.

Our study had limitations. First the population was heterogeneous and small. Second we did not include a control group due to ethical considerations. The 3D-HRAM is able to provide information on the functional anatomy of the sphincter complex after reconstruction for ARM. The clinical significance of the method has yet to be proven.

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