



Manometric findings in relation to functional outcomes in different types of anorectal malformations ^{☆,☆☆,★}



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ABSTRACT

Aims: To compare anorectal manometry (AM) in patients with different types of anorectal malformations (ARMs) in relation to functional outcomes.

Methods: A single-institution, cross-sectional study. After ethical approval, all patients ≥ 7 years old treated for anterior anus (AA), perineal fistula (PF), vestibular fistula (VF), or rectourethral fistula (RUF) from 1983 onwards were invited to answer the Rintala bowel function score (BFS) questionnaire and to attend anorectal manometry (AM). Patients with mild ARMs (AA females and PF males) had been treated with minimally invasive perineal procedures. Females with VF/PF and males with RUF had undergone internal-sphincter saving sagittal repairs.

Results: 55 of 132 respondents (42%; median age 12 (7–29) years; 42% male) underwent AM. Patients with mild ARMs displayed good anorectal function after minimally invasive treatments. The median anal resting and squeeze pressures among patients with mild ARMs (60 cm H₂O and 116 cm H₂O respectively) were significantly higher than among patients with more severe ARMs (50 cm H₂O, and 80 cm H₂O respectively; $p \leq 0.002$). The rectoanal inhibitory reflex was preserved in 100% of mild ARMs and 83% of patients with more severe malformations after IAS-saving sagittal repair. The functional outcome was poor in 4/5 patients with an absent RAIR (BFS ≤ 11 or antegrade continence enema-dependence). Rectal sensation correlated significantly with the BFS.

Conclusions: Our findings support the appropriateness of our minimally invasive approaches to the management of mild ARMs, and IAS-saving anatomical repairs for patients with more severe malformations. Level of evidence: III.

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Internal anal sphincter (IAS)-saving sagittal repair methods, including posterior sagittal anorectoplasty (PSARP) for rectourethral fistula (RUF) in males, and anterior sagittal anorectoplasty (ASARP) for females with vestibular and perineal fistula (VF/PF) are standardized approaches for the anatomical reconstruction of anorectal malformations (ARMs) with a fistulous termination of the bowel outside the external anal sphincter (EAS) complex at our institution and others [1,2]. For mild ARMs with a bowel termination mostly within the EAS such as anterior anus (AA) in females and standard perineal fistula (PF) in males, our approach has been minimally invasive, involving serial dilatations

or conservative follow-up only for AA females, and cutback anoplasty for males with PF [3,4] to achieve a satisfactory passage of stool [5].

We recently published the controlled, long-term bowel functional outcomes by type of ARM for these patients [3,4,6,7]. We found bowel function mostly comparable to matched peers in mild ARMs [3,4], and the majority of patients with more severe ARMs achieved social continence with appropriate aftercare [6,7]. Some degree of functional impairment, however, persisted in approximately 1/3 of females with VF/PF, and in 2/3 of males with RUF [6,7].

This study has aimed to objectively uncover the reasons behind the functional impairments observed, particularly among patients with severe ARMs. The findings of AM by type of ARM require further characterization. We performed manometric evaluation of patients with different types of ARMs after standardized treatments, comparing the findings with patient-reported clinical outcomes. To our knowledge, this is one of the largest single-centre studies of AM in ARM patients to date.

1. Methods

1.1. Patients

After ethical approval, all patients treated at our institution between 1983 and 2006 for AA, PF, VF and RUF were cross-sectionally invited to

Abbreviations: ARM, anorectal malformation; AM, anorectal manometry; AA, anterior anus; PF, perineal fistula; VF, vestibular fistula; RUF, rectourethral fistula; EAS, external anal sphincter; IAS, internal anal sphincter; PSARP, posterior sagittal anorectoplasty; ASARP, anterior sagittal anorectoplasty.

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answer a detailed postal questionnaire on bowel function. All consenting participants ≥ 7 years of age were also invited to undergo anorectal manometry (AM) as outpatients. Patients with major cognitive impairment, severe sacral anomalies (< 3 segments remaining), Currarino syndrome or meningomyelocele were excluded. Participation was voluntary. Operative and case details were obtained retrospectively from records. A single independent investigator conducted the survey and AM. The investigator, an experienced clinician, had not been involved in any aspect of the surgical or medical management of the patients.

1.2. Questionnaires

Bowel function was evaluated using the Rintala bowel function score (BFS) [8], for which a close correlation with clinical outcomes has been established cohorts [8–10], and a large pool of control data is available [11]. A BFS of $\geq 17/20$, achieved by $\geq 90\%$ of controls [11] was taken as the lower limit of normal.

1.3. Manometric equipment and technique

AM was performed using a saline-perfused 4-channel catheter (0.2 ml/min; pressure rise rate 100 cm H₂O/s) with 4 spirally placed side openings (4.5 mm outer diameter) at 1 cm intervals near the tip. Measurements were taken using transducers in each line connected to a personal computer. The machine (Medtronic Polygram 98; Medtronic Functional Diagnostics, Skovlunde Denmark) was calibrated separately for each patient. Patients performed a rectal enema at home the preceding evening. AM was performed without sedation with the patient in the left lateral decubital position with the knees and hips flexed. The lubricated catheter was introduced 10 cm into the rectum and withdrawn by continuous pull-through technique at a rate of 1 mm/s.

1.4. Manometric recordings

The length of the anal canal high-pressure zone (HPZ) was defined as the distance between the proximal margin of the HPZ, indicated by a rise in the anal canal pressure, and the anal outlet characterized by a pressure drop in the distal HPZ to 50% of the maximum [12]. The anal canal resting pressure (ARP) was taken as the mean static pressure when the catheter was placed in the HPZ for 1 min [13]. The anal squeeze pressure (ASP) was taken as the maximum of 3 attempts when the patient was asked to contract their anal sphincter around the catheter [14].

1.5. Rectoanal inhibitory reflex (RAIR) and rectal sensory threshold (RST)

RAIR was provoked by inserting a second catheter containing a latex balloon at a distance of 10 cm from the anal verge and incrementally insufflating the balloon starting from 10 ml of air with the manometry catheter in the HPZ [12–14]. A RAIR was considered to be present if the anal canal pressure dropped by at least 25% of the basal tone over at least 5 s. The rectal sensory threshold (RST) was taken as the mean value of 3 gradual fillings after asking the patient to indicate as soon as they perceived the balloon.

1.6. Reference values for manometric data used in this study

The reported normal values for the HPZ in children and adults have ranged from 2 to 4 cm depending on age [14–17]. For pressure reference values, we used data reported from 17 controls aged 8–17 years who underwent AM at our institution using the same technique: ARP 60 (45–80), ASP 120 (80–184), and 100% for RAIR [18]. AM had been performed by our two senior pediatric colorectal surgeons. As previously reported normal values for RST have ranged been < 15 –20 ml [12,19], > 20 ml was considered indicative of reduced rectal sensation.

1.7. Statistics

Data are presented as median (range). Categorical variables were compared using Fisher's exact test, and continuous variables using the Mann–Whitney *U* test. Spearman's correlation coefficient was used to test the correlation between manometric variables and the functional outcome by BFS. A two-tailed *p*-value < 0.05 was considered statistically significant.

2. Results

2.1. Participants

The main patient characteristics are shown in Fig. 1. Of 132 survey participants aged ≥ 7 years, 55 patients (42%; median age 12 (7–28) years) agreed to AM. All patients had been treated and systematically followed up by the same surgical team from birth and none had been lost to follow-up. Constipation was treated using dietary modifications, laxatives and/or enemas as appropriate. Ten patients (18%) had mild sacral dysplasia (≥ 3 segments remaining), including 0% of females with AA, 1 male with PF (8%), 4 females with VF/PF (19%) and 5 males with RUF (50%).

2.2. Surgical management

All 11 females with AA had been managed non-operatively [3]. Five (45%) had been treated for mild anal stenosis with Hegar dilatations from size 11 to 14. In females, termination of the anal canal mostly within the external sphincter complex (distinguishing AA from PF) was confirmed using an electrical muscle stimulator under anesthesia if this had been clinically unclear [3]. All males with PF had received standard cut-back anoplasty on the first day of life [4]. Females with VF/PF had undergone internal sphincter-saving ASARP [6] with conservation of the distal part of the fistulous bowel termination at a median of 1.1 (range, 0.1–11) months of age; 7 (33%) under colostomy cover. RUF patients (30% bulbar fistula ($n = 3$); 50% prostatic fistula ($n = 5$), and 20% bladderneck fistula ($n = 2$)) had been treated with internal sphincter-saving PSARP [7] after primary colostomy at a median age of 2 (range, 1–10) months. All operatively managed patients had undergone a standard anal dilatation program over 6 weeks up to Hegar size 14, after which any colostomies were closed.

2.3. Postoperative complications and late operations

One male with PF had anoplasty for residual stenosis at 1 year of age. One female with VF required revision of ASARP at the age of 5 years. This patient suffered from intractable constipation, which led to gradual perineal body breakdown. Hirschsprung's disease was histologically excluded. She and two others (1 VF female and 1 RUF male) also underwent resection of a megarectum later in childhood. Three males with RUF (5% of 55 AM participants) aged 9, 10 and 28 years had antegrade continence enema (ACE) conduits for social continence (1 bladderneck and 2 prostatic fistulas).

2.4. Non-participants

Of the 77 patients who had participated in the survey but declined AM (Fig. 1), patient characteristics including gender (56% male), median age (13 range, 7–29 years), and percentage with sacral dysplasia (10%) or ACE conduits (8%) were not significantly different from survey respondents who underwent AM ($p \geq 0.21$ for all comparisons). The types of ARMs (31% AA females, 31% PF males, and 25% RUF males) were also comparable ($p = \text{NS}$), apart from a higher proportion of VF/PF females in the AM group (13% vs 38%; $p = 0.002$). The baseline characteristics of the 63 patients who did not participate any aspect of the study were not significantly different from survey respondents ($p = \text{NS}$ for all comparisons).

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