Long-term bowel functional outcomes in rectourethral fistula treated with PSARP: controlled results after 4–29 years of follow-up

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Abstract

Background/purpose: Posterior sagittal anorectoplasty (PSARP) has become the standard surgical treatment for males rectourethral fistula (RUF) at most surgical centers worldwide. We aimed to define the long-term bowel functional outcomes following PSARP for RUF at our institution between 1983 and 2006, with comparison to age- and gender-matched controls.

Methods: Patients were invited to answer a detailed, previously validated Bowel Function Score (BFS) questionnaire by post. Respondents were matched by age and gender to three controls from the general population who had answered identical questionnaires. Case records were reviewed retrospectively for operative details. Ethical approval was obtained. Social continence was defined as soiling or fecal accidents <1 week and no requirement for changes of underwear or protective aids.

Results: Of 34 (79%) respondents (median age 19 (range, 4–29) years), 74% had voluntary bowel movements (VBMs), 24% were reliant on antegrade continence enema (ACE) washouts, and 1 patient had a colostomy. Impairment of bowel function was significantly higher in all aspects of fecal control among patients than controls (p < 0.001). A statistically significant decline in fecal accidents and soiling was observed with age (p ≤ 0.03). Thirty-one percent of patients with VBMs had constipation managed with diet or laxatives (vs 2% of controls, p = 0.0002). Of patients with VBMs followed up for >12 years (n = 20), 50% were completely continent (vs 73% of controls; p = NS). Overall, 76% of respondents were socially continent with or without artificial means in the form of ACE washouts. By BFS score, 39% had a good functional outcome, 27% had a moderate outcome, 9% had a clearly poor score and 24% were living with an ACE.

Conclusions: Our results suggest that in the long-term, functional symptoms remain highly prevalent among patients treated for RUF with PSARP. However, the majority can be expected to achieve social continence, although for some this will require intervention with ACE bowel management. Approximately one third may report VBMs and complete continence.

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There have been no long-term, controlled studies of functional outcomes focusing solely on males with rectourethral fistula (RUF) treated with posterior sagittal anorectoplasty (PSARP). Most studies of outcomes of anorectal malformations (ARMs) have pooled several types of malformation and modes of treatment, making interpretation of results in relation to individual types of malformation challenging [1–3]. Different series have also used highly variable criteria for the evaluation of fecal continence [4,5], and few studies have included healthy controls [6–11]. PSARP operation, introduced by De Vries and Peña in 1982 [12], is the standard technique for the correction of RUF at most pediatric surgical centers, including ours. PSARP involves division of the anal sphincter and levator muscles in the midline, separation of the rectum and fistula from the urethra, and anatomical repositioning of the rectum in the middle of the sphincter complex [13]. Despite systematic aftercare, the functional prognosis for higher ARMs, such as RUF terminating in the bladder neck and prostate, is less optimistic than for lower lesions such as perineal fistula, owing to greater associated sphincter hypoplasia and motility problems [14,15]. It is thought that rectourethral bulbar fistula, an intermediate type of anomaly in the Wingspread classification [16] is associated with less morbidity than the higher types of urethral fistulae [15]. As imperfections in bowel control also prevail in the general population [17], this study aimed to define the long-term functional outcomes of patients treated with PSARP for RUF in relation to age- and gender-matched controls, to clearly outline where abnormalities in bowel control exist.

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1. Methods

1.1. Patients

All males treated for RUF with sigmoidostomy followed by primary PSARP operation at our institution between 1983 and 2006 were identified from case records and invited to answer a detailed, previously validated bowel function score (BFS) questionnaire [8,18] by post. Patients with total sacral agenesis (n = 1) were excluded. Patients were approached by an independent investigator who had not been involved in their surgical care. Parents assisted children below the age of 16 years in responses. The case records of each patient were retrospectively reviewed for operative details. The ethics committee of the Helsinki University Hospital approved the study protocol.

1.2. Questionnaires

The BFS is an established, 7-item qualitative scoring system developed for the assessment of bowel functional outcomes in benign anorectal disorders [6,8,18,19] for which data from healthy individuals are also available [17]. It can be completed by the child or their parents, and does not require a physical examination [10]. Elements assessed are voluntary control of defecation, rectal sensation, stooling frequency, soiling, constipation and the social impact of these. The maximum score is 20, and scores have been shown to correlate closely with clinical outcomes [6,17,19]. A BFS of ≥ 17, which was achieved by ≥ 90% of healthy individuals aged 4 years and older in our previous study, was taken to be within the normal range and indicative of a good outcome [17]. Parents of patients up to age 12 years were also inquired the age at which diapers for stool were discontinued. The BFS questionnaire is shown in Fig. 1.

1.3. Statistics

Data are given as median (range) or as frequencies. Statistical analysis was performed using Fisher's exact test unless otherwise stated. A p-value of <0.05 was considered statistically significant.

1.4. Controls

Recently, our research group collected a population of 594 subjects aged 4–26 years who had been randomly selected from the Population Register Centre of Finland to serve as controls for children and adolescents with anorectal disorders [17]. From this control population, 3 age- and gender-matched controls were randomly selected for each patient. Two patients, aged 28 and 29 years had to be matched to 26 year-old controls, as these were the oldest individuals available from our pool. All controls had answered identical BFS questionnaires to patients.

2. Results

2.1. Patients

As shown in Fig. 2, of a total of 45 eligible males, two had died. None were lost to follow-up. Thirty-four patients (79%) responded to the questionnaire. The median age of respondents was 19 (4–29) years and all had been followed up since birth. The median age of controls was 18.3 (4–26) years. The fistula was bulbar in 12 (35%), prostatic in 18 (53%) and at the bladder neck in 4 patients (12%). The respondent characteristics by age group are shown in Table 1. There were no patients with major mental retardation. Three patients, aged 6, 10 and 29 years had mild learning difficulties but were included as they had been able to attend school. Nine respondents (26%) had a mildly dysplastic sacrum (3–4 segments remaining). The lateral sacral radiograph was normal in the remainder of respondents.

Twenty-five respondents (74%) had voluntary bowel movements (VBMs) and 8 respondents (24%) only opened their bowels using...
anterograde continence enema \[20\] (ACE) washouts. Three patients in the VBM group had previously had ACE conduits, and one patient, aged 5 years, underwent ACE formation shortly after completing this questionnaire. The median age at ACE formation had been 8 \((5–22)\) years, for which the indication had been deficient fecal control in all cases. One adult respondent had recently undergone temporary colostomy formation owing to persistent diarrhea. The etiology has remained unclear despite extensive investigations, including colonoscopy. Because of the colostomy, only the social item of his questionnaire could be included, leaving 33 complete responses for analysis. However, this patient had been socially continent with VBM and without any form of bowel management up to the onset of watery stools two years previously. Since this study ended, the colostomy has been successfully closed.

2.2. Operative management

Thirty-one (91\%) patients had undergone standard PSARP (with laparotomy in one case) and 3 patients (9\%) with high fistulae had undergone laparoscopic-assisted PSARP. All patients had a covering sigmoid colostomy formed on the first day of life. The median age at PSARP was 3 \((1–18)\) months, which in all cases was performed by a consultant pediatric surgeon. Beginning 2 weeks after PSARP, the neanus was dilated gradually over a period of 6–8 weeks up to Hegar 14, after which the sigmoidostomy was closed. One anal stricture requiring anoplasty occurred during the dilatation period. There were 5 (15\%) stoma-related complications (bowel prolapse \(n = 4\); bowel obstruction \(n = 1\)). The median time from PSARP to stoma closure was 4 \((2–7)\) months.

2.3. Late complications and other anorectal procedures

Rectal strictures occurred in two patients who had undergone PSARP prior to 1991, when rectal tapering was performed if significant dilatation was present at the time of repair (9 taperings in total). The strictures were diagnosed 8 months and 4 years after PSARP and treated with dilatations in one and rectal stricturoplasty in the other. Two patients, born in 1985 and 1996 both required resection of megarectum 4 years after PSARP owing to uncontrolled constipation. One further patient underwent laparoscopic rectectomy for rectal prolapse 3 years after laparoscopic-assisted PSARP. Minor mucosal ectopy requiring local operative correction on 1–2 occasions occurred in 4 patients.

2.4. Nonrespondents \((n = 9)\)

The characteristics of nonrespondents did not differ significantly from those who responded to the questionnaire (median age 20 years; range, 13–27 years). The fistula was prostatic in 8 \((88\%)\) and at the bladder neck in 1 \((12\%).\) All had undergone a standard PSARP operation with covering sigmoidostomy and 2 \((22\%)\) currently had an ACE conduit (Fig. 2).

3. Bowel function

3.1. Fecal control among respondents

To demonstrate overall cross-sectional bowel function outcomes following systematic aftercare, the results of all respondents \((n = 33)\), including those with ACE, were initially considered together. As shown in Fig. 3, impairment of bowel function was significantly higher in all aspects of fecal control among patients than controls \((p < 0.001)\).

3.2. Social continence

In this study, we defined “social continence” as fecal accidents or soiling occurring \(< 1\) week and not requiring changes of underwear or protective aids. Of 33 respondents, 76\% \((n = 25)\) overall were socially continent based on this definition, including 6/8 patients with ACE \((by\ artificial\ means)\), and 19 of the 25 patients with VBM, compared to 96\% of controls; \(p = 0.002\). The percentage of respondents who were socially continent and had VBM was therefore 58\% \((n = 19/33)\).

3.3. Effects of age on soiling and fecal accidents

To study the effects of increasing age on fecal control, patients were divided into two age groups \((\leq 12\) years and 13–29 years\) based on the findings of our previous study \([17]\) showing that the fine-tuning of fecal control may continue to develop up to 12 years of age under normal circumstances. As shown in Fig. 4, there was a statistically significant decline in fecal accidents and soiling by age \((p \leq 0.03)\) among respondents. Frequent impairment \((> 1\) week) was also less common among older patients, but the difference was not statistically significant for either symptom. Social continence by age group was 56\% in patients \(\leq 12\) years of age \((n = 5, including 3 patients with ACE)\) and 83\% in patients aged 13–29 years \((n = 20, including 3 patients with ACE; p = 0.72)\).

3.4. Constipation and bowel management

Of patients with VBM \((n = 25)\), 31\% \((n = 8)\) had constipation managed with dietary or oral laxatives \((vs 2\% of controls; p = 0.0002)\). Bowel frequency was normal in 67\% of these patients \((vs 87\% of controls; p = 0.07)\), more frequent \((3–5\) times per day) in 5 \((21\%)\), and less frequent in 3 \((12\%)\). In the 8 patients with ACE, bowel actions were produced using washouts 3–4 times per week. The proportion of patients requiring adjunctive measures to produce satisfactory bowel actions by age group is shown in Fig. 5. The mean age at ACE formation had been 5 \((4–5)\) years in the 4–12 years age group, and 12 \((7–22)\) years in patients aged 13 and older. The percentage of respondents free from any form of intervention \((dietary, oral laxatives or ACE washouts)\) was significantly higher above age 12 compared to patients \(\leq 12\) years of age \((70\% vs 11\% respectively; p = 0.005)\), suggesting improvement in function over time.

3.5. Outcomes by BFS

As shown in Fig. 6, 67\% \((n = 22/33)\) of respondents had a BFS consistent with a good \((BFS \geq 17)\) or moderate \((BFS 12–16)\) outcome. The outcome was poor \((BFS \leq 11)\) in 9\% \((n = 3)\) and one quarter were
living with an ACE. The median BFS among patients with VBMs was 17 (mean 16.1 ± 3.8; 6–20) compared to 19 in controls (mean 19.1 ± 1.7; 11–20; p = 0.0001; Mann–Whitney U-test). The 3 patients with a poor BFS include two patients aged 5 years, one of whom has since had an ACE procedure and is now successfully toilet-trained, and one adult in his late twenties who still uses protective aids for social reasons but has not opted for an ACE. He was among the first patients to undergo PSARP procedure (bulbar urethral fistula) at our institution.

3.6. Outcomes in patients with VBMs

Of the 25 patients in the VBM group, 43% (n = 11) had difficulties withholding defecation until an appropriate time (vs 5% of controls; p < 0.0001), 28% (n = 7) had impaired sensation of the need to defecate (vs 9% of controls; p = 0.04), 60% (n = 15) soiled (vs 29% of controls; p = 0.005), and 48% (n = 12) had fecal accidents (vs 8% of controls; p < 0.0001). Ten patients (40%; all >12 years of age) had no soiling or fecal accidents (vs 69% of controls; p = 0.02) and of these, 5 (20% of 25 patients) had a BFS of 20/20 (vs 59% of controls; p = 0.002). The symptom profile of patients with VBMs followed up for more than 12 years (n = 20) is shown in Fig. 7. Problems withholding defecation and fecal accidents remained significantly higher than in controls even after long-term follow-up (p < 0.005), and frequent soiling was more prevalent than in controls (15% vs 2% in controls; p = 0.04). However, 50% (n = 10) were free from soiling and fecal accidents (vs 73% of their respective controls; p = 0.096), including 3 patients with diet- (n = 2) or laxative-managed (n = 1) constipation but normal bowel frequency. Additionally, 85% (17/20) of respondents in this group were socially continent, including the 5 patients who had reported no bowel symptoms at all (BFS 20/20).

![Fig. 3. Impairment of fecal control among patients (n = 33) vs controls (n = 99).](attachment:image.png)

![Fig. 4. Effects of age on soiling and fecal accidents among respondents (n = 33).](attachment:image.png)
3.7. Outcomes by level of fistula

The effects of the level of fistula on the proportion of patients who had VBMs, the median BFS and the percentage who were completely continent for stool are shown in Table 2. Of the 4 patients with bladder neck fistulae, after a follow-up period of 16–27 years all soiled occasionally and 50% still experienced fecal accidents. However, 3/4 of these patients were socially continent (including 2 patients with ACE).

3.8. Toilet training for stool

Of the 9 patients aged 4–12 years, 7 had successfully completed toilet training for stool at a median age of 5 (2.5–7.3) years (vs 2.3 (1.3–4.0) years in controls; \( p < 0.0001 \) using the unpaired t-test). In 5 cases, this had only occurred after an ACE procedure (one ACE had already been closed). Two patients (22%) had completed toilet training for stool without intervention at 2.5 and 5 years, and 2 patients, both aged 5, still wore diapers at times owing to fecal accidents or soiling. One of these patients has since had an ACE conduit formed and has since discontinued wearing diapers completely.

3.9. Social problems

Of 33 responses, the prevalence of social problems was 36% (n = 12) compared to 5% of controls (\( p < 0.0001 \)). These restricted social activities in two (6%) and were severe enough to cause psychological disturbance in one case (3%), occurring “sometimes” only in the remaining 27%. The prevalence was 38% (3/8) in patients with ACE and 36% (9/25) in the VBM group (\( p = \text{NS} \) between groups). There was no significant difference in the prevalence of social problems by age group (33% in patients up to 12 years of age (n = 3, of which 2 “sometimes” and 1 restricting social life), and 37% in those older than 12 years (n = 9, of which 7 “sometimes”, 1 restricting social life and 1 severe); \( p = \text{NS} \) between age groups). The patient with a temporary colostomy also reported social problems restricting social life, which were mainly owing to the watery stools that have since settled.

4. Discussion

During the last 30 years, PSARP has become the operation of choice for high ARMs such as RUF, replacing classical pull-through procedures [21–23]. In this study we have aimed to systematically define the functional outcomes of a single type of malformation treated with a standardized procedure, which has included comparison with healthy controls. The questionnaires were sent and reviewed by an independent investigator who has not been involved in the surgical care of these patients, which should encourage patients to report their symptoms freely. As no respondent had total sacral agenesis or major mental retardation, this reduces the number of confounding factors involved, allowing consideration of results principally in relation to RUF treated with PSARP. Consistent with other series [24], major PSARP-related complications with a potential impact on bowel control were uncommon.

A cross-sectional analysis including all 33 responses (Fig. 3) showed that individual aspects of fecal control were significantly inferior among RUF patients in relation to matched controls (\( p < 0.001 \)). The prevalence of any degree of impairment of function ranged from 35% for rectal sensation to 70% for soiling. The figure for soiling is similar to, but lower than the 82% reported by Rintala and Lindahl (1995) [8] in 46 patients with high and intermediate anomalies (23 males with RUF). Bliss and coworkers (1996) [25] also reported absence of soiling in only 23% of 53 patients treated for high imperforate anus with PSARP (35 males with RUF). The inclusion of females with cloaca and patients with major sacral anomalies in both these studies may explain the higher rates of soiling observed. As ACE conduits form an integral part of the modern management of a significant proportion of RUF patients, these results outline the overall continence outcomes that are achieved with systematic aftercare in this patient population as a whole.

Encouragingly, our data suggest that frequent impairment (> 1/week) involving any particular symptom only affects between 9 and 18% of RUF patients in the mid to long term (Fig. 3). We also observed a significant
decline in the prevalence of both soiling and fecal accidents with time (Fig. 4), suggesting that functional status improves with age. This observation has also been made in other studies of high anorectal malformations after both PSARP [7,24] and classical procedures [26–28]. It is unlikely that true improvement in sphincter function occurs. It is more likely that there is some adaptation to residual dysfunction and improved overall cognition of the problem. However, the benefits of vigorous and early management of functional complications, especially constipation on outcomes are also well established [8,24,29–31].

The prevalence of constipation managed with diet or laxatives among patients with VBMs in our series was 31% (n = 8). Including those patients with ACE (n = 8), 48% (16/33) overall required adjunctive measures to produce satisfactory motions, which concurs closely with 42% reported by Hassett et al. (2008) [32] in the same subset of patients. The change in the prevalence of constipation and requirement for bowel management by age group (Fig. 4) is likely to represent an improvement in constipation over time, while also reflecting a trend towards earlier intervention with ACE. Although our observations are limited by the small number of patients ≤12 years of age (n = 9), other studies have also found subsiding constipation around puberty [7,28,33].

A cross-sectional analysis of all respondents by BFS showed that a good functional outcome (BFS ≥17) was achieved by 39% of patients, a moderate outcome by 27%, and 9% had a clearly poor score (Fig. 5), consistent with other studies [8,29]. Additionally, one quarter of respondents in our series were presently living with an ACE. On one hand, they could be taken to represent patients with the poorest outcomes. However, the fact that the majority are socially continent with washouts (6/8 patients; 75%) makes their outcome seem altogether more favorable. ACE conduits also enabled completion of toilet training for stool in 2/3 of those patients from whom this was enquired. In reports, 2/3 to 96% of patients gain continence with an ACE [22,34], and this procedure is both reversible and often psychologically more acceptable to patients than repeated rectal enemas. Our current practice has become to offer an ACE conduit to all RUF patients with deficient continence from the age of 4 years onwards to enable discontinuation of diapers prior to commencing elementary school.

Voluntary bowel movements (VBMs), defined as feeling an urge to defecate, the capacity to verbalize this feeling, and the ability to hold the bowel movement [35] have been used as an outcome criterion in studies of ARMs. In Peña’s series (1995) [29], 64% (79/123) of RUF patients had VBMs following PSARP, and this figure was 79% (27/34) in the follow-up study by Bliss and co-workers (1996) [24] and 74% by cross-section in ours if all patients are included (Fig. 2). Although VBMs imply adequate functioning of the rectum, pelvic floor and sphincter apparatus for fecal control [32,36], defining what constitutes normal fecal continence remains a challenge. Occasional soiling in particular, appears to affect one half to one quarter of otherwise healthy children and young adults in an age-dependent manner, and fecal accidents also occur in 5% overall [17]. Frequent soiling or accidents (>1/week) requiring changes of underwear or protective aids, however, are key features that are uncommon among controls (4% of 99 controls in this study) and formed the basis of our definition for “social continence.”

Further to reporting the results of all patients with VBMs (n = 25), we specifically analyzed outcomes in VBM patients who had been followed up for >12 years (n = 20). By this time, any improvement in bowel function is likely to have reached an end-point and any residual dysfunction is likely to be permanent. While problems withholding defecation and fecal accidents remained significantly higher than in controls even after this follow-up period (Fig. 7), most symptoms were occasional (<1/week) only. Of these 20 patients, 85% (n = 17) were socially continent; 50% (n = 10) were completely continent (no soiling or fecal accidents) and a quarter (n = 5) reported no bowel

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**Table 2**

<table>
<thead>
<tr>
<th>Level of fistula</th>
<th>n (%)</th>
<th>VBM (%)</th>
<th>Median BFS in VBM patients (range)</th>
<th>No soiling or fecal accidents (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bulbar</td>
<td>12 (38%)</td>
<td>11 (92%)</td>
<td>18 (6–20)</td>
<td>5 (42%)</td>
</tr>
<tr>
<td>Prostatic</td>
<td>17 (52%)</td>
<td>13 (76%)</td>
<td>16 (7–20)</td>
<td>5 (29%)</td>
</tr>
<tr>
<td>Vesical</td>
<td>4 (12%)</td>
<td>1 (25%)</td>
<td>15 (15)</td>
<td>0 (0%)</td>
</tr>
</tbody>
</table>

* p<0.005 compared to controls; p=NS for other symptoms vs controls

**Fig. 7.** Fecal control in patients with VBM followed up for >12 years (n = 20).
symptoms at all (BFS 20/20). Therefore, of the 24 patients in this series who had been followed up for >12 years (including 4 patients with ACE), the rate of complete continence and VBM was 42% (n = 10). As no patient ≤12 years of age was entirely free of soiling and fecal accidents, this equates to rates of complete continence and VBM in 30% (10/33) of our respondents with RUF.

In a recent study, Hassett and colleagues (2009) [32] reported continence rates of 58% in 19 RUF patients after 10 years of follow-up, but our results for complete continence are closer to the 35% reported by Rintala and Lindahl (1999) [21], and 36% in Peña’s series (1995) [29], which is the largest to date. However, as only 73% of our controls >12 years of age were completely free from any soiling and fecal accidents, it is important to consider the figures in patients in relation to this. It may be that social continence, achieved by 96% of our controls, represents a measure for comparison that is achieved by almost all healthy individuals. In perspective, of 33 RUF patients, 76% (n = 25) in total were socially continent with or without artificial means in the form of ACE washouts. However, only 58% (19/33) had VBM and social continence, based on their responses.

Of the 24 patients followed up for >12 years, 83% in total (n = 20/24) were socially continent under normal circumstances, including 17 of the 20 patients who opened their bowls voluntarily (85%), and 3/4 of the patients who opened their bowls using ACE washouts. Therefore, in the long term, 71% of patients (17/24) had achieved VBM and social continence. Considering that in previous reports only 5–7.5% of children operated using classical methods were reported to have achieved complete continence [24,28,37], the outcomes of ours and other studies nonetheless unequivocally support the superiority of PSARP for the treatment of high anomalies. The important role of ACE conduits in securing social continence for this subset of patients is also emphasized.

By level of fistula, our data support the conclusions of others [7,25,29] that higher urethral fistulae carry a worse functional prognosis (Table 2), although a similar proportion of patients with both bulbar and prostatic urethral fistulae had achieved VBM. While social problems owing to bowel function were reported by 36% of respondents, all but 9% occurred only and these did not cluster specifically among those respondents with ACE or to any particular age group. The negative social consequences of poor bowel function have been described from a number of perspectives [23,26,30] and minimizing social disability in patients with ARMs remains a fundamental objective of clinical management.

5. Conclusions

This report has aimed to systematically define the long-term functional outcomes of patients with RUF following PSARP operation in relation to healthy controls. Our results suggest that while functional symptoms are highly prevalent among patients, at least 2/3 will develop VBM, and approximately 30% will report complete continence. However, the majority of patients can be expected to achieve social continence following systematic aftercare, although for some this will require intervention with ACE bowel management. While our results support the notion that a degree of functional improvement may occur over time, the role of social adaptation in this process remains to be defined. The effective management of constipation and timely intervention with adjunctive measures such as ACE conduits remain essential for achieving social continence in those who would otherwise have poor control. Toilet training for stool is likely to be delayed in comparison to peers, and parents should be informed of this. As our results for fecal continence are in keeping with those of other investigators, this report may provide a useful basis for the management and prognostic counseling of patients with RUF and their families. The long-term outcomes of PSARP operation for RUF have not been previously described in such a detail including comparison with healthy controls.

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References


