

# Sexual Function, Fertility and Quality of Life after Modern Treatment of Anorectal Malformations



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**Purpose:** Sexual dysfunction and impaired quality of life due to fecal incontinence are common after classic operations for anorectal malformations. We hypothesized that modern repairs may result in improved outcomes.

**Materials and Methods:** Following ethical approval for this single institution cross-sectional study, all patients 16 years or older treated for rectourethral, vestibular or perineal fistula from 1983 onward were sent detailed postal questionnaires on sexual function and quality of life. Each respondent was age and gender matched to 3 controls randomly selected from the general population. Penoscrotal/gynecologic abnormalities were obtained from the records.

**Results:** A total of 41 patients (62%) with a median age of 22 years participated in the study. Of the patients 20 were males with rectourethral fistula (prostatic in 60%), 10 were females with vestibular/perineal fistula and 11 were males with low malformations. Although experience of sexual relationships and orgasmic function were reported in comparable proportions to controls, age at coital debut was significantly delayed in all groups of patients ( $p \leq 0.046$ ). Erectile function was preserved after sagittal repair but absent ejaculations or azoospermia affected 3 males with rectourethral fistula (15%). Penoscrotal/gynecologic abnormalities affected 12% of patients. Overall quality of life scores were comparable to controls but a trend was found for lower scores on emotional items in males with rectourethral fistula ( $p = 0.06$ ) and for a negative effect on sexual life in females ( $p = 0.03$ ).

**Conclusions:** While erectile and orgasmic function appear preserved after sagittal repair, further evaluation of fertility issues in males with rectourethral fistula is indicated. Larger multicenter studies are needed to confirm our findings.

**Key Words:** colorectal surgery, fertility, fistula, quality of life, sexual development

THE introduction of posterior sagittal anorectoplasty in 1982 and its later modification, anterior sagittal anorectoplasty involving repair of anorectal malformations under direct vision, have significantly improved the functional outlook for individuals compared to patients undergoing a classic pull-through operation.<sup>1-3</sup> Modern bowel management programs, including antegrade continence enema

conduits, have allowed achievement of social continence among the majority of cases, including those involving severe anorectal malformations.<sup>1,2</sup> Patients with mild anorectal malformations can largely expect a functional outcome comparable to matched peers by adulthood with appropriate aftercare.<sup>4-6</sup>

Patients treated during the PSARP era have now reached an age at which

## Abbreviations and Acronyms

ACE = antegrade continence enema  
ARM = anorectal malformation  
ASARP = anterior sagittal anorectoplasty  
EHS = Erection Hardness Score  
GIQLI = Gastrointestinal Quality of Life Index  
PF = perineal fistula  
PSARP = posterior sagittal anorectoplasty  
QoL = quality of life  
RUF = rectourethral fistula  
VF = vestibular fistula

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evaluation of the wider impact of the ARM on their lives is possible. Sexual function, personal relationships and the ability to have children are important determinants of QoL, impairment of which was common after classic repairs in patients with ARMs.<sup>7,8</sup> Similarly the negative effects of fecal incontinence on QoL and social functioning, which affected a significant proportion of patients in the past, are well established.<sup>9,10</sup> More recent series examining sexual function following classic repairs have resulted in conclusions consistent with earlier studies.<sup>7,8,11,12</sup> In contrast to classic methods, PSARP involves approaching the fistulous bowel termination under direct vision using a strict midline approach, which theoretically reduces the risk of iatrogenic damage to the delicate structures involved in reproductive function, particularly in males. We evaluated controlled outcomes for sexual function and QoL in patients with ARMs treated exclusively during the era of sagittal repairs to determine the extent to which they are associated with improved outcomes.

## METHODS

### Patients

This was a single institution cross-sectional study. After ethical approval all patients 16 years or older treated from 1983 onward for ARMs were invited to answer a postal questionnaire on sexual function and QoL. Those evaluated included males with RUF, females with vestibular or perineal fistula and males with low ARMs (PF/anal stenosis). Questionnaires were sent up to 3 times. An independent investigator conducted the survey. Major cognitive impairment (3 patients), total sacral agenesis and Currarino syndrome (2) were exclusion criteria. Case records were reviewed retrospectively. Three individuals matched for age and gender were randomly selected for each patient from a previously collected reference population of 594 individuals randomly selected from the Population Register of Finland to serve as controls for patients with ARMs.<sup>13</sup> Controls answered identical questionnaires to patients. Three patients 28 to 29 years old were matched to 26-year-old controls since they were the oldest available from our pool. Three controls per patient was the maximum number available from our pool.

### Questionnaires

Relationship status, age at coital debut and orgasm during intercourse (usually/sometimes/seldom/never) was inquired of all respondents. Erectile function was rated based on the validated Erection Hardness Score (range 0, penis does not enlarge, to 4, penis is hard and fully rigid).<sup>14</sup> Ejaculations and age at spermarche were inquired of males, and age at menarche of females. Patients were asked whether they had tried to have children, number of pregnancies/children fathered and any fertility issues/treatment.

### Quality of Life

QoL in adulthood was evaluated from all respondents 18 years or older using the validated GIQLI (maximum score 144 points).<sup>15</sup> Scores lower than 105 are indicative of constant gastrointestinal symptoms.

### Statistics

Data are presented as median (range) unless otherwise stated. Responses of patients and controls were compared. Categorical variables were evaluated using the Fisher exact test and continuous variables with the Mann-Whitney U test, with  $p < 0.05$  being considered significant.

## RESULTS

Patient characteristics are outlined in table 1. Of 66 eligible patients 41 responded. Of 20 males with RUF 30% had bulbar, 60% prostatic and 10% bladder neck fistulas. A total of 11 males had low ARMs (PF or anal stenosis) and 10 females had VF/PF. No patient was lost to followup. Eight patients (20%) had mild sacral dysplasia (3 to 4 segments remaining). QoL data were collected from all respondents 18 years or older (35 patients). Median age of the 123 controls (93 males) was 22 years (range 16 to 26), which did not differ significantly from patients.

### Operative Management by ARM Type

All males with RUF underwent PSARP under colostomy cover at a median age of 3 months (range 2 to 24). All females with VF or PF underwent ASARP at a median age of 3.5 months (range 0.1 to 48), of whom 50% had a covering colostomy. Males with low ARMs had undergone cutback anoplasty and/or anal dilation on the first day of life except 2 patients

**Table 1.** Respondent characteristics

	RUF Group	Low ARM Group	VF/PF Group	Overall
No. gender/total No. (%)				41/41 (100)
Male	20/41 (49)	11/41 (27)	0/41 (0)	
Female	0/41 (0)	0/41 (0)	10/41 (24)	
Median yrs age (range)	22 (16–29)	21 (16–29)	21 (16–28)	21 (16–29)
No. voluntary bowel movements/total No. (%)	18/20 (90)	11/11 (100)	10/10 (100)	39/41 (95)
No. ACE/total No. (%)	2/20 (10)*	0/11 (0)	0/10 (0)	2/41 (5)
No. socially continent/total No. (%)†	17/20 (85)	11/11 (100)	10/10 (100)	38/41 (93)

Patients with RUF underwent PSARP, those with low ARMs underwent anoplasty/anal dilation and those with VF/PF underwent ASARP.

\* Both patients had bladder neck fistulas.

† Less than 1 soiling or fecal accident weekly (no requirement for protective aids, ie pads).

referred late, in whom anoplasty was performed at age 1 year. Patients underwent a standard 6-week anal dilation program up to Hegar size 14 beginning 2 weeks postoperatively.

### Associated Genital/Gynecologic Abnormalities

Five patients (12%) had genital/gynecologic anomalies. Four males with RUF (20%) had penoscrotal anomalies, of whom 3 had bifid scrotum, 2 had hypospadias and 3 had testicular maldescent (bilateral in 2). Among females there were no gynecologic abnormalities apart from pyosalpinx in a teenage girl, in whom salpingectomy was performed. We routinely perform a check colposcopy at puberty and abdominal ultrasound of the gynecologic organs at least once during followup.

### Nonrespondents

Among 25 nonrespondents 8 males (32%) had RUF, 12 males (48%) had low ARMs and 5 females (20%) had VF/PF. There were proportionally but statistically insignificantly more males with low ARMs among nonrespondents ( $p = 0.11$ ). Operative treatment by ARM type, length of postoperative followup, age (median 21 years, range 16 to 28), gender (80% male), ACE requirement (8%), sacral dysplasia (16%) and penoscrotal/gynecologic abnormalities (16%) were comparable to respondents ( $p \geq 0.72$ ).

### Gross Bowel Function in Respondents

At survey 39 patients (95%) had voluntary bowel movements, and 2 with bladder neck fistulas (10% of males with RUF) used ACE washouts and lacked voluntary control. A total of 38 subjects (93%), including 17 of 20 with RUF, were socially continent with or without bowel management (less than 1 soiling or fecal accident weekly and no requirement for protective aids).<sup>3</sup>

### Sexual Function in Males

Similar proportions of male patients and controls had engaged in sexual intercourse ( $p \geq 0.08$ ) but coital debut was delayed in males with RUF and males with low ARMs compared to controls (table 2).

Normal orgasms were noted in the majority of males with RUF and in all males with low ARMs. Of the 2 males with RUF who experienced orgasms at least some of the time 1 had fathered 2 children. Erectile function was normal (EHS 4) or sufficient for penetration (EHS 3) in all males with RUF. Of the 2 patients with EHS 3 a prostatic fistula was present in 1 and a bladder neck fistula in 1. No respondent reported an EHS below 3.

Ejaculations were absent in 2 males, 18 and 23 years old, with RUF. One had a bladder neck fistula and had undergone repair of bifid scrotum and penile hypospadias as well as unilateral orchiectomy for an atrophic testis during childhood (EHS 3), and 1 had undergone prostatic RUF repair, had normal erections (EHS 4) and had no penoscrotal abnormalities. Two males with RUF (6% of male patients and 7% of male controls,  $p = 1.00$ ) did not respond to this item. Spermarche had occurred normally at a median age of 13 years (range 10 to 16) in the males with RUF who responded to this item, and in all of those with low ARMs and controls ( $p = 0.43$ ).

Of the 4 males with penoscrotal anomalies 1 (25%) was in a stable relationship and 2 (50%) had engaged in sexual intercourse, including 1 of the 2 patients with hypospadias. Of the 2 patients with bladder neck fistulas and ACE conduits (ages 18 and 27 years) the older patient was in a stable relationship and engaged in sexual intercourse but not until age 24 years. All 4 males with mild sacral dysplasia had normal erections (EHS 4) and ejaculations.

### Sexual Function in Females with VF/PF

There was no significant difference in the proportion of females in a stable relationship or with experience of intercourse ( $p \geq 0.27$ , table 3). However, as with males, coital debut was 2 years later than in controls (18 vs 16 years,  $p = 0.046$ ). Orgasms were comparable to controls. Menarche had occurred in 9 of 10 patients at a median age of 13 years (range 11 to 14) and in all controls at age 13 years (11 to 16,  $p = 0.26$ ). One patient had primary amenorrhea at age 18 years but normal gynecologic anatomy and is under

**Table 2.** Controlled outcomes for sexual function in males

	RUF Group			Low ARM Group		
	Pts	Controls	p Value	Pts	Controls	p Value
No. stable relationship/total No. (%)	6/20 (30)	30/60 (50)	0.19	1/11 (9)	13/33 (40)	0.08
No. sexual intercourse/total No. (%)	10/20 (50)	41/60 (68)	0.18	6/11 (55)	21/33 (64)	0.72
Median yrs age at coital debut (range)	18 (15–24)	16 (13–24)	0.04	19 (14–20)	16 (12–22)	0.04
No. normal orgasm during intercourse/total No. (%)*	8/10 (80)	39/41 (95)	0.09	6/6 (100)	20/21 (95)	1.00
No. normal erections/total No. (%)†	18/20 (90)	58/60 (97)	0.26	11/11 (100)	32/33 (97)	1.00
No. normal ejaculations/total No. (%)	16/18 (89)‡	55/55 (100)‡	0.06	11/11 (100)	31/31 (100)‡	1.00

Patients with RUF underwent PSARP and those with low ARMs underwent cutback/anal dilation.

\* Normal orgasm usually/most of the time.

† EHS 4.

‡ Two patients and 5 controls did not respond to item.

**Table 3.** Controlled outcomes for sexual function in females with VF/PF after ASARP

	Pts	Controls	p Value
No. stable relationship/total No. (%)	3/10 (30)	17/30 (50)	0.27
No. sexual intercourse/total No. (%)	8/10 (80)	26/30 (87)	0.63
Median yrs age at coital debut (range)	18 (14–20)	16 (14–20)	0.046
No. normal orgasm during intercourse/total No. (%)*	6/10 (75)	20/30 (77)	1.00
No. normal menarche/total No. (%)	9/10 (90)	30/30 (100)	0.25

\* Orgasm usually/most of the time.

endocrinological investigation. Of 4 females with mild sacral dysplasia the 3 who were sexually active had normal orgasms.

### Fertility

Only 2 males (both with RUF, ages 27 and 29 years, with bulbar and prostatic fistulas) had attempted to become parents. Both reported normal ejaculations. One patient had fathered 2 children, and 1 with azoospermia and infertility had required bilateral orchiopexy and hypospadias repair during childhood.

### Quality of Life

Overall GIQLI scores were comparable between patients and controls ( $p \geq 0.09$ , table 4). Statistically insignificantly more patients with RUF and VF/PF had total scores less than 105 (17% and 13%, respectively, vs 4% in controls,  $p \geq 0.09$ ). Among subscores (scale 0 to 4, with 4 indicating no effect at all<sup>15</sup>) a negative effect on sexual life was observed only in females with VF/PF (median 3 points vs 4 points in controls,  $p = 0.03$ ). A trend toward uncontrolled stools was also observed in females (median 3.5 vs 4 points in controls,  $p = 0.07$ ). Males with RUF tended to score lower in coping with everyday stresses (median 3 vs 4 points in controls,  $p = 0.06$ ). In males with PF QoL was comparable to controls.

## DISCUSSION

We evaluated the sexual function outcomes, fertility status and GIQLI for patients with ARMs treated during the era of modern sagittal repairs. Matched controls were used to define areas of abnormality. We achieved a good response rate (62%), with 50% of the cohort comprising males with severe ARMs, for whom sexual function outcomes after PSARP are

of great interest. Essential characteristics between respondents and nonrespondents were comparable in the dropout analysis (difference not significant). Gross fecal control, which impacts social functioning, was relatively good by adulthood. The majority of patients achieve reasonable social continence after sagittal repairs,<sup>1,4</sup> which is superior to results with traditional techniques.<sup>16–18</sup>

There was no significant difference in the proportion of patients in stable relationships compared to controls, or in the proportion who had engaged in sexual intercourse overall or by type of ARM (table 2). This finding suggests acceptable social integration even in patients with severe ARMs. However, later coital debut ( $p \leq 0.046$  vs controls) suggests some effect from the ARM on willingness to commence intimate relationships, and this finding has been reported by others.<sup>12</sup> Nonetheless, the proportions of males and females with ARMs who had engaged in sexual intercourse by their 20s were largely similar in this and other studies.<sup>12,19,20</sup> Coital debut among controls corresponded with population based data from developed countries.<sup>12,21</sup>

Nearly all males had normal erections (EHS 4), including 18 of 20 (90%) with RUF following PSARP. Only 2 males reported an EHS of 3, which is nonetheless sufficient for penetration. This finding compares favorably to significant erectile dysfunction in 21% to 30% of patients in earlier series, mostly after traditional pull-through operations.<sup>12,20</sup> Orgasms were also preserved after PSARP, being normal in 8 of 10 males with RUF and occurring at least some of the time in the remaining 2 (of whom 1 had fathered children normally). Hence, damage to the pelvic nerves may be considerably less likely after PSARP than classic operations due to the use of direct vision.<sup>11,22,23</sup> No sexual dysfunction occurred in males with low ARMs after simple anal cutback. Although PSARP is performed in these patients at some centers, there are no studies to support its superiority over simple cutback, and urological injuries have been described.<sup>23</sup>

Normal ejaculations occurred in 16 of 18 males with RUF (89%) and in all males with low ARMs (table 2). Ejaculations were absent in 2 patients with RUF and 1 patient had azoospermia despite apparently normal ejaculations, comprising 15% of males

**Table 4.** GIQLI scores by ARM type

	RUF Group	Low ARM Group	VF/PF Group	Overall
Pts:				
Median score (IQR)	124 (115–133)	136 (134–137)	122 (113–134)	132 (120–136)
No. score below 105/total No. (%)*	3/18 (17)	0/9 (0)	1/8 (13)	4/35 (11)
Controls:				
Median score (IQR)	132 (123–135)	133 (128–137)	125 (120–132)	131 (124–135)
No. score below 105/total No. (%)	2/18 (4)	0/9 (0)	1/8 (4)	3/35 (3)

\*  $p \geq 0.09$  vs controls for all groups.

with RUF. Previously abnormalities affected 17% to 41% of patients with intermediate and high ARMs after various surgeries.<sup>11,12,20</sup> Although intraoperative injuries to reproductive structures (divided vas, opened seminal vesicle) are reported in only 0.3% to 2% of cases after PSARP, it is likely that their actual incidence, particularly during dissection of high fistulas, is greater.<sup>22–24</sup> Ejaculate analysis is needed to determine the extent of the problem.

Sexual function, including orgasmic function, after ASARP for VF/PF in females was not significantly different from controls (table 3). Similar data for orgasms (65% vs 70% in our study) but a high incidence of dyspareunia (63%) has been reported in a previous study that included patients with cloaca.<sup>12</sup> We did not inquire about a history of dyspareunia, and in retrospect these data would have been useful, although tissue dissection in ASARP is considerably less compared to full PSARP or cloacal repair, thus minimizing scarring. Another major weakness of our study is the small number of patients by type of ARM due to the relative rarity of the condition. This factor introduces the possibility of type II error, ie there may have been differences that were undetected due to small sample size. However, our cohort comprises predominantly severe ARMs (50% were males with RUF and 24% were females with VF/PF), in whom problems with sexual function and QoL might be expected to cluster. Assessment of patients with cloaca is part of our future work. Although assessment of sexual function was not based on a formally validated questionnaire, matched controls demonstrate how the general population responds to these items. QoL and erectile function were assessed with validated surveys.

Apart from 1 pyosalpinx the absence of structural gynecologic abnormalities such as vaginal septum, which affects 5% of females with VF, is likely to be coincidental. Gynecologic abnormalities occur in up to 20% of females with VF,<sup>25</sup> but are uncommon in those with PF.<sup>26</sup> In males hypospadias, bifid scrotum and undescended testicles are typical associations of RUF.<sup>24</sup> Testicular maldescent, affecting 3 patients with RUF (15%) in this series, reportedly occurs in 27% of those with high and 7% of those with low ARMs.<sup>27</sup>

Due to the small number of patients, assessment of the effects of penoscrotal abnormalities on sexual

relationships was limited. However, of the 4 patients with penoscrotal abnormalities 2 had commenced sexual relationships, which is comparable to the overall patient cohort. Similarly 1 of the 2 patients with the most severe ARMs (bladder neck fistula) and ACE washout dependent continence was in a stable sexual relationship, although coital debut was 8 years later than the median in controls (16 years). Mild sacral dysplasia (3 or more segments remaining) had no demonstrable effect on sexual function.

Regarding fertility, only 2 males with RUF had attempted to become parents, of whom 1 had fathered 2 children and the other was infertile. In the infertile patient the azoospermia could relate to blocked vasa either congenitally or from corrective surgery for the ARM, or from bilateral orchiopexy or epididymitis. Among females with ARMs successful pregnancies are not uncommonly reported.<sup>11,12,25</sup> In 1 series there was no difference in fertility rates between females with ARMs and the general population.<sup>20</sup> Obstetric surveillance is advisable, and we advocate delivery by cesarian section since considerable residual defects in the pelvic floor may be present.<sup>28</sup>

Compared to controls, no significant differences in overall GIQLI scores were observed, although a negative effect on sexual life ( $p = 0.03$ ) was observed among females as well as a trend toward uncontrolled stools ( $p = 0.07$ ), and males with RUF tended to report more difficulty coping with daily stresses ( $p = 0.06$ ). In females a fear of fecal incontinence in particular has been associated with depressive symptoms, whereas in males these symptoms are predicted by perceived difficulties in forming personal relationships.<sup>29,30</sup> Thus, the possible effects of ARMs on QoL may be subtle and difficult to uncover, and targeted psychological interventions should consider possible gender differences.

## CONCLUSIONS

In contrast to patients undergoing ARM repair with classic methods, most domains of sexual function in our patients appeared comparable to controls. Importantly erectile and orgasmic function appeared preserved after PSARP for RUF but ejaculatory abnormalities may continue to affect a proportion of patients. Larger multicenter studies and evaluation of fertility issues are needed.

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## EDITORIAL COMMENT

The authors describe sexual function, fertility and quality of life outcomes after treatment for anorectal malformations. This topic is important and has not previously been well studied. Given that the cohort assessed was a relatively small, heterogeneous group, further study is still necessary. Also given the patient ages, fertility outcomes were limited. However, the data presented certainly add to available information about long-term outcomes in patients with anorectal malformations.

A noteworthy strength of this investigation includes the survey response rate achieved (62%), which is much higher than is typically seen. This

high response rate may be a function of the health care system in which the investigators practice and may be difficult to replicate in other settings. Developing generalizable, efficient strategies for long-term followup of the large group of pediatric surgical patients who may have altered sexual function and fertility should be a priority for our field.

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