

Outcome of patients with anorectal malformations after posterior sagittal anorectoplasty: a study from Ahvaz, Iran

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Aim and purpose The aim of this study was to evaluate the outcome of patients who underwent posterior sagittal anorectoplasty (PSARP) for the treatment of low or high anorectal malformation (ARM).

Patients and methods All patients who underwent standard PSARP were included in this study. Patients with mental retardation were excluded from our study. Patients were classified according to the Rintala score into four categories: poor (6–9); fair (9–11); good (12–17); and normal (18–20). We used a questionnaire introduced by Rintala. The type of anomaly was divided into two categories. We used low and high ARM definitions according to the relationship of the terminal colon to the levator muscles of the pelvic floor. The Student *t*-test, the Pearson χ^2 -test, one-way analysis of variance, and the Levine test were used for data analysis using SPSS ver. 13.0.

Results Sixty patients aged 3–17 years (13.63 ± 3.27 years) were included. The mean of score in patients with low-type ARM was 14.5 ± 2.6 and that in patients with high-type ARM was 13.19 ± 3.75 ($P=0.28$). The mean of scores was 13.34 ± 3.5 among male patients and 13.94 ± 2.9 among female patients. There was no statistically

significant difference ($P=0.46$). The score was significantly higher in patients with fistula ($n=51$, 13.9 ± 3.1) than in patients without fistula ($n=9$, 11.8 ± 3.3 ; $P=0.03$).

Excluding two cases with scrotal-type fistula and rectal atresia, there was no significant difference between the two groups ($P=0.06$).

Conclusion There was no significant difference in the outcome after PSARP between boys and girls. There was no significant difference between low-type and high-type ARM. The mean of score was significantly higher among patients with fistula than among patients without fistula. *Ann Pediatr Surg* 10:65–67 © 2014 Annals of Pediatric Surgery.

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Introduction

Results of anoplasty can be evaluated by objective and subjective methods. The objective method is less favorable now because this method is expensive and may be unavailable, especially in developing countries. Objective methods include manometry, computed tomography, MRI, anal sphincter myography, and intra-anal sonography [1]. There are several scoring systems suggested by Kelly [2], Templeton and Ditesheim [3], Kiesewetter and Chang [4], Holschneider [5], and Rintala and Lindah [6]. The Kelly score [2] requires rectal examination by hand and the Holschneider [5] score requires anorectal manometry. The Rintala score does not require physical examination and is a purely subjective method [6]. The aim of this study was to evaluate the outcome and quality of life among patients with low or high anorectal malformation (ARM) who underwent posterior sagittal anorectoplasty (PSARP).

Patients and methods

Sixty patients (male 32, female 28) who underwent standard PSARP were included in this study. These patients underwent surgery during the past 1–15 years. Patients with mental retardation were excluded from our study. We used low and high ARM definitions according

to the relationship of the terminal colon to the levator muscles of the pelvic floor [7]. After surgery, parents were educated on the dilatation program. Dilatation was performed for each patient by parents according to the age of the patient after 2 weeks of repair. The protocol of dilatation used for patients is shown in Table 1 [8]. The outcome of cases was assessed using the validated bowel function score (Table 2).

Patients were classified according to their scores into four categories: poor (6–9); fair (9–11); good (12–17); and normal (18–20). We used a questionnaire introduced by Rintala and Lindah [6]. Voluntary control, sensation, soiling, stool frequency, constipation, and the social impact of constipation were assessed by the Rintala score [9]. The Student *t*-test, the Pearson χ^2 -test, one-way analysis of variance, and the Levine test were used for data analysis using SPSS (ver. 13.0; SPSS Inc., Chicago, Illinois, USA). The person who filled the questionnaire was not a member of the medical team. This study was approved by the ethical committee of the university.

Results

Sixty patients aged 3–17 years (13.63 ± 3.27 years) were included in this study (Table 3). Of the patients, 32

Table 1 The dilatation protocol used in our patients

Age	Hegar dilator
1–4 months	12
4–8 months	13
8–12 months	14
1–3 years	15
3–12 years	16
≥ 12 years	17

Table 2 Bowel function scores

Factors	Scores
Urge to defecate	
Always	3
Most of the times	2
Sometimes/uncertain	1
Never	0
Ability to hold back defecation	
Always	3
Problems (<1 per week)	2
Problems weekly	1
Never	0
Frequency of defecation	
Twice a day to every other day	2
More often	1
Less often	1
Soiling	
Never	3
Occasional (<1 per week)	2
Frequent (>1 per week)	1
Daily	0
Constipation	
Never	3
Manageable with diet	2
Manageable with laxatives	1
Manageable with enema	0
Social problems	
No	3
Sometimes (foul odors)	2
Restricting social life	1
Severe	0

Table 3 Distribution of age of patients included in the study

Age	<i>n</i>
3	1
5	3
6	3
7	1
9	4
10	8
11	5
12	4
13	14
14	7
15	6
17	3
Total	60

(53.33%) were male and 28 (46.67%) were female ($P = 0.46$). Of them, 29 patients (48.4%) were 3–12 years old and 31 patients were more than 12 years old. The mean of age was 10.77 ± 2.94 years. Out of 60 patients, 29 (48.33%) had low anomaly and 31 (51.67%) had high anomaly ($P = 0.71$).

The mean of scores among all patients was 13.63 ± 3.27 . The minimum and the maximum of scores were 7 and 18,

Table 4 Average of scores among different types of cases with or without fistula

Type of fistula	<i>n</i>	Mean \pm SD
No fistula	9	11.89 ± 3.37
Perineal	15	14.67 ± 2.22
Urethral	6	14.00 ± 3.95
Vaginal	9	15.11 ± 2.31
Vestibular	9	14.44 ± 2.29
Vesical	10	11.80 ± 4.18
Scrotal	1	16 ^{NA}
Rectal atresia	1	7 ^{NA}

NA: Mean \pm SD was not applicable.

Table 5 Prognosis among patients with high or low anorectal malformation

	Poor	Fair	Good	Normal
High anomaly [<i>n</i> (%)]	5 (16.12)	6 (19.35)	17 (54.83)	3 (9.70)
Low anomaly [<i>n</i> (%)]	1 (3.45)	4 (13.80)	21 (72.41)	3 (10.34)
Total [<i>n</i> (%)]	6 (10)	10 (16.67)	38 (63.33)	6 (10)

$P = 0.33$.

respectively. The mean of scores in patients with low-type ARM was 14.5 ± 2.6 and in patients with high-type ARM was 13.19 ± 3.75 . There was no significant difference between the two groups ($P = 0.28$).

The mean of scores was 13.34 ± 3.5 among male patients and 13.94 ± 2.9 among female patients. There was no statistically significant difference ($P = 0.46$).

There was no significant correlation between the score and the age of the patients (Pearson coefficient = 0.13). The score was significantly higher among patients with fistula ($n = 51$, 13.94 ± 3.19) than among patients without fistula ($n = 9$, 11.8 ± 3.3 ; $P = 0.03$). Excluding two cases with scrotal-type fistula and rectal atresia, there was no significant difference between the two groups ($P = 0.06$). The average of scores among different types of ARMs is shown in Table 4. The minimum (score = 7) score was seen in a patient with rectal atresia and the maximum (score = 16) was seen in a patient with scrotal fistula (Table 4).

Poor prognostic cases were seen in 16.12% of patients with high ARM and 3.45% of patients with low ARM ($P = 0.22$). Of 31 patients with high anomaly, 17 (54.83%) had good prognosis. Of 29 patients with low anomaly, 21 (72.41%) had good prognosis ($P = 0.15$, $\chi^2 = 1.99$). There was no significant difference between high and low ARM regarding their prognosis evaluated by the Rintala score (Table 5).

Fecal soiling and some degree of fecal incontinence were seen in 67 and 31.7% of the cases, respectively.

Discussion

In our study, the male/female ratio was 1.14/1. In the study from South Africa, the male/female ratio was 1.6/1 [10]. In the study from Pakistan, of 100 neonates with ARM, 77 were male and 23 were female (male/female = 3.4/1) [11]. Hence, boys are affected more often than girls in these studies.

In the study carried out by Goyal *et al.* [12], the best prognosis was reported in cases with perineal fistula and poor prognosis was seen in vesical fistula. These findings were similar to our study. In the study by Hassett *et al.* [13], the best prognosis was reported in cases with perineal followed by vestibular and urethral fistula. In their study, a posterior sagittal approach was used for treatment. A questionnaire was used for the scoring system.

In the study of Kaselas *et al.* [14], the highest score was seen in patients with perinanal fistula. In our study, the highest score was seen in cases with vaginal fistula (15.1 ± 2.3) followed by perineal fistula (14.6 ± 2.3). Vesical fistula had the poorest prognosis in the study of Kaselas *et al.* [14] and in our study.

Rintala *et al.* [9] compared children with low ARM with normal healthy children. They concluded that only half of the children with a low ARM have age-appropriate normal bowel function.

In our study, there is no significant score improvement with increasing age. In some studies, as age increased, there was improvement in their score [9].

Hassink [15] conducted a study on patients with low-type anomaly. Of the patients, 83% showed good prognosis and 15% had fair and poor prognosis. Our findings are similar. In the study of Hassink, there was a positive correlation between prognosis improvement and increasing age [15]. In our study, there was no significant correlation between age and prognosis improvement.

In our study, the most common complication after surgery was constipation. Other studies also reported constipation as the major postoperative complication in patients who underwent PSARP [16,17]. The incidence of constipation has been reported to be from 10 to 73% in patients who underwent PSARP [16,18]. In the study of Rintala *et al.* [9], constipation was present in 42% of the patients.

In our study, 31.7% of the patients had fecal incontinence. Elhalaby [19] reported that incontinence was present in 33.3% of their 38 patients. In the study of Elhalaby, 18 out of 38 patients were more than 3 years old. In our study, all patients were more than 3 years old. Improvement in constipation in patients with increasing age was reported by Rintala and Linadhl [20]. Hence, constipation is expected to resolve in our patients when they reach adolescence. In the study by Ibrahim [21], of 23 neonates whose follow-up periods were longer than 3 years, 21 neonates had a good score using the Kiewewetter score [4].

Conclusion

Constipation is higher in our study than in other studies. The number of patients with normal sphincteric function is lower than in other studies. There was no significant difference between boys and girls. There was no significant difference between low-type and high-type

ARM. The mean of scores was significantly higher among patients with fistula than among patients without fistula. Careful follow-up and parent education are also recommended to achieve a better outcome.

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Conflicts of interest

There are no conflicts of interest.

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