

Original Research Article

The analysis of continence after posterosagittal anorectoplasty in anorectal malformation patients in Wahidin Sudirohusodo hospital of Makassar

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ABSTRACT

Background: The management of anorectal malformations universally uses posterior sagittal anorectoplasty (PSARP) as standard surgery. The aim of this research was to determine continence in patients with anorectal malformation after PSARP action.

Methods: This study was a descriptive analytical categorical research with cross sectional design. The data were obtained from patients' medical records and questionnaire given to patients underwent PSARP in Wahidin Sudirohusodo Hospital and network hospitals from June 1 to December 1, 2018. The analysis was done to 60 children ranging from 5 to 8 years old. Most of them were male (56.7%) ranging from 5 to 6 years old (53.3%).

Results: The results of the research indicated that based on malformation subtype, most of them suffered from rectourethra fistula (40.0%) with continence problem (61.7%). There was no significant correlation between sex and continence problem ($p > 0.05$). However, it was seen that the percentage of continence subjects was higher in female (65.4%) than in male (58.8%), while the percentage of soiling and constipated subjects was higher in male than in female. Rectourethra fistula was found more frequently in male than in female (70.6%), while vestibular fistula was found more frequently in female (69.2%). There was no significant correlation between age and defecation problem ($p > 0.05$). However, the percentage of continence and soiling subjects was higher in 5-6 years children then the one for 7-8 year children, while the percentage of constipated subjects was higher in 7-8 year children (21.4%) than 5-8 year children (18.8%).

Conclusions: Gender differences in the outcome of children with anorectal malformations must be considered. Men with perineal fistula were likely to experience continence and constipation than women with perineal fistulas. Women with perineal fistulas and vestibular fistulas had almost similar outcome.

Keywords: Anorectal malformation, Continence PSARP, Defecation disorder, Fistula type, Krickenbeck classification

INTRODUCTION

Anorectal malformation is a congenital abnormality without anus or with imperfect anus, including agenesis of ani, agenesis of rectal, and rectal atresia. This

abnormality could be accompanied by VACTERL syndrome (Vertebrae, Anal, Cardial, Esophageal, Renal, Limb).^{1,2} Children with anorectal malformations have a variety of spectrum disorders rectosigmoid motility. Patients with anorectal malformations who underwent

surgery with maintained rectosigmoid, generally experiencing constipation.¹ This is possible due to the hypomotility of the rectosigmoid area in patients with anorectal malformations. Therefore, constipation is more common in patients with low and moderate anorectal malformations in children. Anorectal malformations of rectosigmoid loss would experience the opposite.³⁻⁵

Anorectal malformations vary widely, from the low-position imperforate anus where the rectum is in a normal location but too narrow so the baby's stool could not pass through, intermedia anorectal malformations where the rectum is close to the urethra and high location anorectal malformations where the anus is completely absent.⁴

The average incidence of anorectal malformations worldwide is 1 in 5000 births.⁶ In general, anorectal malformations are more common in men than in women. The rectourethral fistula most commonly found in male infants, followed by perineal fistulas. Whereas in female infants, the most common types of anorectal malformations are rectovestibular fistula and perineal fistula.⁷

Anorectal malformations have a long history. For centuries, experts have been trying to improve babies with this abnormality.³ Babies that survive generally are babies who have low localized malformation abnormalities. In 1835, Amussat was the first to do suturing between the rectal mucosa and the edge of the skin. This action was thought to be the first act of actual anoplasty. In the first sixty years of the twentieth century, many anoplasty procedures were performed on low-lying abnormalities. Since the introduction of the colostomy technique, colostomy has been carried out in a high-lying malformation disorder.⁸

In the era of abdominoperineal pull through surgery, surgery was carried out by pulling the rectum as close to the sacrum as possible to avoid injury to the urogenital tract. Stephens in 1953 introduced a sacred approach with the aim of preserving puborectal slings which is an important factor in maintaining continence. The posterosagittal approach was introduced in 1980. This approach turned out to provide a great operating area exposure, therefore this technique then widely used today.⁸

Posterior Sagittal Anorectoplasty (PSARP) facilitates better exposure to the complex sphincter muscle and through the incision in the posterior part of the midline, therefore the new rectum could be placed in the right place.⁵

Previous research by Goyala et al, conducted functional research after surgery for anorectal malformations using the Rintala scoring system. In the study, the average functional outcomescore (FOS) was 13.7 in male patients and 14 in female patients. FOS progressively worsens with the severity of anorectal malformations.¹

Data on the evaluation of the defecation function after PSARP that carried out in Makassar has not found yet. By estimating the PSARP in the Wahidin Sudirohusodo hospital about 20-30 patients for each year, it is necessary to do research to determine the function of defecation after PSARP. The aim of this study was to determine continence in patients with anorectal malformations after PSARP.

METHODS

This research was categorical descriptive analytic with cross sectional design. The study was conducted at Wahidin Sudirohusodo hospital and it's networking hospital. The population were all the patient who underwent PSARP at Wahidin Sudirohusodo hospital and it's networking hospital from June 1st 2018 - December 1st 2018. The samples those met the inclusion and exclusion criteria was taken.

Inclusion criteria

- Patients with a diagnosis of anorectal malformation.
- Patients who underwent PSARP surgery at the Wahidin Sudirohusodo hospital and networking hospital.
- Have sufficient data in the medical record
- Patients available to contacted
- Willing to be included in the study
- With age more than 5 years old

Exclusion criteria

Patients were excluded from the study if they did not visit the hospital to be followed up after underwent PSARP. The instrument used in PSARP continence based on previous research conducted by Krickenbeck (Krickenbeck Classification).

There were two types of data in this study, secondary data from medical records to complete the existing scoring questionnaire and primary data regarding the function of defecation after surgery obtained from interviewed by phone or patient's visit at the hospital pediatric surgery in Wahidin Sudirohusodo hospital.

Statistical analysis

Bivariate correlation analysis was used to determine the frequency distribution (proportion) of demographic characteristics and the component of continence scoring of respondents (stated significant when $p < 0.05$). Data was processed using SPSS 22.

RESULTS

During the study period there were 60 pediatric patients with age range from 5 years to 8 years, consisting of 34 men and 26 women. Malformation subtype distribution was found to be significantly different between men and

women ($p < 0.001$). Rectourethra fistula was found more common in men than women with percentage 70.6%. Whereas vestibular fistulas are more common in women with percentage 69.2% (Table 1). The distribution of anorectal malformation subtype by age did not differ significantly ($p > 0.05$). It was seen that perineal fistula

and rectourethra had similar percentage 37.5% and was higher than vestibular fistula 25.0% in 5-6 years group, while rectourethra fistula with percentage 42.9% was higher than perineal fistula 21.4% and vestibular fistula 35.7% in 7-8 years group (Table 2).

Table 1: Distribution of malformation subtypes by gender.

| Gender | Malformation subtype | | | Total | P |
|--------|----------------------|--------------------|----------------------|-------|-------|
| | Perineal fistula | Vestibular fistula | Rectourethra fistula | | |
| Men | N | 10 | 0 | 24 | 0.000 |
| | % | 29.4% | 0.0% | 70.6% | |
| Women | N | 8 | 18 | 0 | |
| | % | 30.8% | 69.2% | 0.0% | |
| Total | N | 18 | 18 | 24 | |
| | % | 30.0% | 30.0% | 40.0% | |

Table 2: Distribution of anorectal malformation subtypes by age.

| Age | Anorectal malformation subtype | | | Total | P |
|----------|--------------------------------|--------------------|----------------------|-------|-------|
| | Perineal fistula | Vestibular fistula | Rectourethra fistula | | |
| 5-6 year | N | 12 | 8 | 12 | 0.375 |
| | % | 37.5% | 25.0% | 37.5% | |
| 7-8 year | N | 6 | 10 | 12 | |
| | % | 21.4% | 35.7% | 42.9% | |
| Total | N | 18 | 18 | 24 | |
| | % | 30.0% | 30.0% | 40.0% | |

Table 3: Distribution of defecation disorders by gender.

| Gender | Malformation subtype | | | Total | P |
|--------|----------------------|---------|--------------|-------|-------|
| | Continenence | Soiling | Constipation | | |
| Men | N | 20 | 7 | 7 | 0.846 |
| | % | 58.8% | 20.6% | 20.6% | |
| Women | N | 17 | 4 | 5 | |
| | % | 65.4% | 15.4% | 19.2% | |
| Total | N | 37 | 11 | 12 | |
| | % | 61.7% | 18.3% | 20.0% | |

Table 4: Distribution of defecation disorders by age.

| Age | Malformation subtype | | | Total | P |
|----------|----------------------|---------|--------------|-------|-------|
| | Continenence | Soiling | Constipation | | |
| 5-6 year | N | 20 | 6 | 6 | 0.967 |
| | % | 62.5% | 18.8% | 18.8% | |
| 7-8 year | N | 17 | 5 | 6 | |
| | % | 60.7% | 17.9% | 21.4% | |
| Total | N | 37 | 11 | 12 | |
| | % | 61.7% | 18.3% | 20.0% | |

There was no significant association between men and women with defecation disorder ($p > 0.05$), but it was seen that the percentage of continents was higher in

women (65.4%) (OR 1.322) than in men (58.8%) (OR 0.756), while the percentage of subjects who were soiling

and constipated was higher in men than in women (Table 3).

There was no significant relationship between age and defecation disorder ($p > 0.05$), but the percentage of continents and soiling subjects was higher at 5-6 years of age compared to 7-8 years of age (OR = 1.1818), while the percentage of subjects who were constipated was higher at the age of 7-8 years (21.4%) (OR 0.846) than at the age of 5-6 years (18.8%) (Table 4).

There was no significant correlation between the malformation subtype and defecation disorder ($p > 0.05$). The highest percentage of subjects suffering from continence was perineal fistula (83.3%), while the highest percentage of subjects suffering from soiling was rectourethra fistula (29.2%) and the highest percentage of subjects suffering from constipation in the vestibular fistula (27.8%) (Table 5).

Table 5: Correlation between malformation subtype and defecation disorder.

| Malformation subtype | | Malformation subtype | | | Total | P |
|----------------------|---|----------------------|---------|--------------|--------|-------|
| | | Continence | Soiling | Constipation | | |
| Perineal fistula | N | 15 | 0 | 3 | 18 | 0.101 |
| | % | 83.3% | 0.0% | 16.7% | 100.0% | |
| Vestibular fistula | N | 9 | 4 | 5 | 18 | |
| | % | 50.0% | 22.2% | 27.8% | 100.0% | |
| Rectourethra fistula | N | 13 | 7 | 4 | 24 | |
| | % | 54.2% | 29.2% | 16.7% | 100.0% | |
| Total | N | 37 | 11 | 12 | 60 | |
| | % | 61.7% | 18.3% | 20.0% | 100.0% | |

DISCUSSION

A The study showed that there was no significant relationship between sex with impaired defecation ($p > 0.05$), with subjects having continence higher in women (65.4%) than in men (58.8%). These in line with the study of Stenstrom et al, which found that voluntary control of bowel movements was greater in women (75%) than in men (33%). Statistically, voluntary bowel movements between women (70%) and men (58%) were similar. Among children with perineal fistula, 79% of women reported voluntary bowel movements compared to 91% of men. Nearly 75% of women with vestibular fistulas experienced voluntary bowel movements, but only 33% in men with rectourethral fistulas. The frequency of the absence of voluntary bowel movements was similar between women with perineal fistula and women with vestibular fistula, whereas in men with rectourethral fistula the incidence of the absence of voluntary bowel movements was more significant than in men with perineal fistulae.^{7,8}

There was no significant correlation between the malformation subtype and impaired defecation ($p > 0.05$). The highest percentage of subjects suffering from continence was perineal fistula (83.3%), while the highest percentage of subjects suffering from soiling was rectourethra fistula (29.2%) and the percentage of subjects suffering from the highest constipation in the vestibular fistula (27.8%).

In this study there was no significant relationship between age and impaired defecation, but the percentage

of subjects who experienced continence and soiling was higher at the age of 5-6 years compared with those aged 7-8 years. While the percentage of subjects who were constipated was higher at the age of 7-8 years than at the age of 5-6 years. The study of Kayima et al, said that children who were operated at the age of 3 years had better continence results.^{9,10} Constipation was an often finding found after surgical repair of anorectal malformations, occurring in approximately 50% of patients. The reasons for constipation were remain unclear and appear to occur in similar numbers regardless of the surgical technique used.¹¹⁻¹³

The frequency of fecal incontinence was comparable between women (48%) and men (42%). Women with perineal fistula reported had a higher frequency incontinence than men with perineal fistula (42% and 10%) and almost the same as women with vestibular fistulae (50%). Men with perineal fistula had a lower significance of incontinence compared to men with other ARM subtypes.¹⁰

When all incontinence grades (0-3) compared to women, there were no significant differences in incontinence rank distribution when comparing perineal fistulas and vestibular fistulas ($p > 0.3$, Kruskal-Wallis with post hoc test for data ranking). In line with this, severe incontinence was significantly more frequent in subgroups with recto-urethral fistulas and no fistulas compared with men with perineal fistulas.^{10,13}

In patients with perineal fistula, the frequency of severe incontinence was more significant in women than in men.

The prevalence of severe incontinence did not differ between young and old age groups, both for women and men with ARM.¹⁴

The frequency of constipation was significantly higher in women (62%) than men (35%). There was no difference in frequency between children with perineal fistula and other ARM subtype groups, both in women and men. In patients with perineal fistula, constipation was more common in women than in men. The frequency of constipation was almost similar between women with perineal fistulas and patients with vestibular fistulas.¹³

The rank of consumption level (Krackenbeck grade 0 - 3) did not differ between perineal fistulas and other ARM subtypes ($p > 0.3$ Kruskal-Wallis post hoc test for data ranking). In the prevalence of grade 3 constipation (without diet or laxative management) there was no difference between women and men with perineal fistula ($p > 0.3$, Fisher's exact post hoc test) or between patients with other perineal fistulas and ARM subgroups in each sex ($p > 0.3$ for each comparison, Fisher's exact post hoc test). There was no significant difference in the frequency of constipation between women with ARM who were younger and older. However, the frequency of constipation in men was lower in the older age group than in the younger age group. The loss of voluntary bowel movements did not differ between low and high age groups in men and women.^{10,11}

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REFERENCES

1. Goyal A, Williams JM, Kenny SE, Lwin R, Baillie CT, Lamont GL, et al. Functional outcome and quality of life in anorectal malformations. *J Pediatr Surg.* 2006;41(2):318-22.
2. Gangopadhyay AN, Pandey V. Anorectal Malformations. *J Indian Assoc Pediatr Surg.* 2015;20(1):10-5.
3. Oldham K, Colombani P, Foglia R, Skinner M. Principles and Practice of Pediatric Surgery Vol.2. Philadelphia: Lippincott Williams & Wilkins, 2005;1395-1434.
4. Boocock G, Donnai D. Anorectal Malformation: Familial Aspects and Associated Anomalies. *Arch Dis Childhood.* 1987;62:576-9.
5. Levitt MA, Pena A. Anorectal Malformations. In: Coran AG, Adzick NS, Krummel TM, Laberge JM, Shamberger RC, Caldamone AA, eds. *Pediatric Surgery.* 7th ed. Philadelphia, PA: Elsevier Saunders; 2012:1289 -1309.
6. Grosfeld J, O'Neill J, Coran A, Fonkalsrud E. *Pediatric Surgery* 6th edition. Philadelphia: Mosby elsevier, 2006;1566-1599.
7. Oldham K, Colombani P, Foglia R, Skinner M. Principles and Practice of Pediatric Surgery Vol.2. Philadelphia: Lippincott Williams & Wilkins, 2005;1395-1434.
8. Pena A. Anorectal Anomalies. *Pediatric Surgery, Diagnosis and Management.* 2009;463-76.
9. Kayima P, Kit D, Punchak M, Andersen GA, Situma M. "Patterns and treatment outcomes of anorectal malformations in Mbarara Regional Referral Hospital, Uganda." *J Pediatr Surg.* 2018:21-5.
10. Kella N, Memon S, Qureshi G. Urogenital Anomalies Associated with Anorectal Malformation in Children. *World J Med Sci.* 2006;1(2)151-4.
11. Van den Hondel D, Sloots CE, Gischler SJ, Meeussen CJ, Wijnen RM, I Jsselstijn H. Surgical Long-term Follow Up team. Prospective long-term follow up of children with anorectal malformation: Growth and development until 5 years of age. *J Pediatr Surg.* 2013;48:81825.
12. Hassett S, Snell S, Hughes-Thomas A, Holmes K. 10-Year outcome of children born with anorectal malformation, treated by posterior sagittal anorectoplasty, assessed according to the Krackenbeck classification. *J Pediatr Surg* 2009;44:399-403.
13. Pakarinen MP, Koivusalo A, Lindahl H, Rintala RJ. Prospective controlled long-term follow up for functional outcome after anoplasty in boys with perineal fistula. *J Pediatr Gastroenterol Nutr.* 2007;436-9.
14. Stenström P, Kockum CC, Benér DK,. Adolescents with anorectal malformation: physical outcome, sexual health and quality of life. *Int J Adolesc Med Health.* 2014;26(1):49-59.

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