Patient's outcome after definitive surgery using krickenbeck continence score in anorectal malformation

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ABSTRACT

Introduction: Anorectal malformations (ARM) are a common type of congenital anomaly seen in pediatric surgery, with an approximate incidence ranging from 1 in 2,000 to 5,000 live births. The main aim of neonatal care in patients with ARM is to ensure that patients achieve optimal functionality after definitive surgery. The Krickenbeck continence score is used to assess the outcomes of ARM patients. So, this study aimed to evaluate patient outcomes after definitive surgery in Arifin Achmad General Hospital.

Materials and Methods: This was a cross-sectional retrospective study utilizing electronic medical records of patients with ARM who had undergone a definitive surgery procedure at Arifin Achmad General Hospital, Indonesia, from 2019 to 2022.

Results: In this study, twenty-six patients with ARM were identified, including 12 males and 14 females. Approximately 76.9% of the patients had normal birth weight. The study found that 61.5% of the patients had ARM without fistula, 15.4% had ARM with rectourethral and perineal fistula, 3.8% had ARM with vestibular fistula, and 3.8% had ARM with cloaca. According to the Krickenbeck continence score, 61.5% of the patients achieved Voluntary Bowel Movement (VBM), while 11.5% experienced soiling and 7.7% experienced constipation. It was observed that normal birth weight patients had finer VBM compared to those with low birth weight (P <0.001), and male patients had finer VBM than females (P = 0.005).

Conclusion: The results of ARM patients for functional outcomes at our hospital are generally positive. More than half of the children exhibit voluntary bowel movements (VBM), and only a little experience soiling and constipation. The number of VBM may be linked to the child's birth weight and gender.

KEYWORDS:
Anorectal malformation, Krickenbeck classification, outcome

INTRODUCTION

Anorectal malformations (ARM) are the most frequent of the gastrointestinal tract congenital anomalies encountered in pediatric surgery practice, consisting of structural anomalies of the rectum, anal sphincter complex, and genitourinary apparatus.[1–3] The reported rate is between 1 in 2,000 and 5,000 live births with no apparent sex predilection.[4,5] Although no defined etiology of ARM has been established, genetic predispositions as well as environmental factors have been thought to be causative.[1] These malformations occur during embryogenesis,[6] characteristically because of urorectal septum arrested caudal migration to the cloacal membrane,[2] or else because of pathologic recanalization processes occurring in the ninth week of gestation, resulting in ectopia of the anal canal in the cloacal structure.[7]

The diagnosis of ARM is primarily established by the observation of meconium or feces passing through ectopic sites. In males, this would be from the perineum or urethra, and in females, this may be observed to pass through the vestibule or perineum. When there is no passage of stool, radiographic evaluation with a prone cross-table lateral view can be indicated to aid in anatomical localization.[8] The Krickenbeck classification system categorizes ARM into three broad domains: diagnostic, surgical management, and long-term functional outcomes following definitive surgery. [9] The main diagnostic subtypes are perineal fistula, rectourethral fistula (with bulbar and prostatic subtypes), rectovesical fistula, rectovestibular fistula, cloacal malformation, absence of fistula, and anal stenosis.7[-10] Rectourethral fistulas are most commonly found in males, whereas rectovestibular fistulas are more commonly found in females.[1,9]

The management of ARMs is based on anatomical classification and the degree of the defect.11 The initiated technique described by Peña and DeVries, the posterior sagittal anorectoplasty (PSARP), has been a very successful procedure. But management is more complex in patients with a poorer prognosis for continence.[8,12–14] Fecal incontinence, in addition to being a physiological problem, is also a significant psychosocial issue, impacting a child's sense of autonomy and subjective quality of life.[6,15] Current surgical goals have evolved beyond ensuring the simple survival of infancy, with a greater emphasis being placed on providing acceptable long-term bowel function to enhance the child's daily living and social integration.[4,6,16,17] Functional outcomes are evaluated by three parameters, as distinguished by Krickenbeck: voluntary bowel movements (VBM), soiling, and constipation.[2,5] This study will determine ARM patient functional outcomes through the Krickenbeck continence scoring system.

MATERIALS AND METHODS

This retrospective study aimed to evaluate the functional outcomes of patients with ARM treated with posterior sagittal

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Table I: Association between the characteristics of ARM patients and functional outcomes

Characteristic	N (%)	VBM		p-value	Soiling		p-value	Constipation		p-value
		Yes	No		Yes	No		Yes	No	
Gender										
▪ Male	12 (46.2%)	11	1	0.005	1	11	1.00	0	12	0.483
▪ Female	14 (52.8%)	5	9		2	12		2	12	
Birth Weight										
▪ Normal	20 (76.9 %)	16	4	<0.001	2	18	1.00	2	18	1.00
▪ Low	6 (23.1 %)	0	6		1	5		0	6	
ARM Type										
▪ No Fistula	16 (61.5%)	9	7	0.741	2	14	0.065	1	15	0.012
▪ Rectoperineal Fistula	4 (15.4%)	2	2		0	4		0	4	
▪ Rectourethra Fistula	4 (15.4%)	3	1		0	4		0	4	
▪ Rectovestibular Fistula	1 (3.8%)	1	0		0	1		0	1	
▪ Cloaca	1 (3.8%)	1	0		1	0		1	0	

anorectoplasty (PSARP) or minimal anoplasty in the Pediatric Surgery Department of Arifin Achmad General Hospital from December 2019 to December 2022. The inclusion criteria were patients aged ≥3 years who had completed all surgical treatment stages at least six months before follow-up, allowing for valid continuity assessment. Participants who had comorbid conditions or who declined to take part in the evaluation were excluded. All the eligible patients received adequate postoperative follow-up to measure the outcome.

This study investigated correlations of gender, birth weight, type of anorectal anomaly, and postoperative functional outcomes. Anomalies were categorized according to the Krickenbeck classification system. Functional outcomes assessed after definitive surgical intervention were voluntary bowel movements (VBM), soiling, and constipation. Voluntary bowel movements were defined as perception of the urge to defecate, communication of need, and control over defecation. Soiling was graded into three levels: grade 1, intermittent soiling (1–2 episodes a week); grade 2, daily soiling with no effect on social interaction; and grade 3, chronic soiling with social handicap. Constipation was graded as well: grade 1, manageable with diet adjustment; grade 2, necessitating pharmacologic therapy with laxatives; and grade 3, not responsive to either diet change or laxatives.[18]

Categorical variables were presented as frequencies and accompanying percentages. Association between the categorical variables was ascertained through Fisher's exact test. A p-value of less than 0.05 was considered statistically significant. IBM SPSS Statistics version 27.0 was used for statistical analysis.

RESULTS

A total of 26 patients with ARM were admitted to Arifin Achmad General Hospital. Among them, 12 patients (46.2%) were male and 14 (53.8%) were female. Most of the patients (76.9%) had a normal birth weight. According to Krickenbeck's classification, the most common anomaly was ARM without fistula, occurring in 61.5% of cases. It was followed by rectourethral and rectoperineal fistulas (15.4% each), vestibular fistula (3.8%), and cloacal malformation (3.8%).

Voluntary bowel movements (VBM) with adequate sphincter control were achieved in 61.5% of patients. Some patients, however, experienced soiling, which happened in 11. 5% of cases, and others had constipation, which was seen in 7. 7% of patients. Normal birth weight patients had significantly greater rates of VBM compared to low-birth-weight patients (p<0.001). Male patients also had significantly improved functional outcomes compared to female patients (p=0.005). On the other hand, the type of anorectal malformation did not make a statistically significant difference in whether or not a patient could achieve voluntary bowel movements (p=0.741).

Additionally, between gender, birth weight, or ARM type, there was no correlation with the occurrence of soiling and constipation, with p-values of 1.0, 1.0, 0.065, and 0.483, 1.0, 0.012, respectively (Table I).

DISCUSSION

Anorectal malformations are a range of congenital anomalies that can vary from mild anal misplacement with a good functional outcome to serious malformations that present significant surgical and management challenges.[9,19] Cassina et al. hold that ARM occurs in about 1 per 5,000 live births, making them comparatively common among congenital abnormalities. Unlike the 2:1 male predominance reported by Houben et al., in the present study, the gender distribution was equal.[1] This result aligns with findings from Mfinanga et al. and Narad et al., who also reported female predominance among ARM patients.[5,15]

Pediatric surgeons taking care of patients with ARM not only for the surgical treatment but also for the follow-up care to achieve bowel continence. All but a few patients with ARM experience some form of defecation dysfunction, and it can significantly impact their quality of life (QOL). Children with ARM have much poorer bowel function than children of similar ages. The extent of functional impairment directly correlates with ARM severity and long-term effects of constipation and/or fecal incontinence, frequently extending into adulthood.[7,13,16]

Krickenbeck classification was developed to standardize the measurement of functional outcomes across different subgroups of surgery so that comparisons between groups are

more meaningful. Bowel function is the most important predictor of long-term functional outcome in children with ARM because fecal incontinence and chronic constipation are serious postoperative complications. These may seriously undermine the social and psychological health of the affected.[6,9] In measuring functional outcomes during assessment, certain critical parameters must be determined: continence, the voluntary control of defecation without episodes of soiling; bowel regularity, more typically described as the frequency of absence of irregular or hard stools; and quality of life in general.[9]

Fecal continence is maintained by a blend of voluntary sphincter control, normal anorectal sensation, and orderly bowel motility. Certain anatomical and clinical characteristics have been shown to function as predictors for excellent functional outcomes in ARM patients. These include normal sacral and spinal anatomy, developed gluteal cleft, presence of anal dimple, ARM types, and the lack of presacral or sacral masses. Despite the presence of all these favorable features, the majority of patients ultimately develop defecatory dysfunction, such as constipation and/or incontinence, as permanent sequelae.[2,7] "True incontinence" has frequently been linked with sacral agenesis and rudimentary pelvic neuromuscular development.[20] Sacral agenesis has an estimated prevalence of between 1 in 25,000 to 100,000 live births, with a rate of 1 in 350 live births occurring among mothers with diabetes mellitus. This condition disrupts neural control, resulting in muscle weakness and sphincter dysfunction, which can lead to issues such as fecal incontinence and constipation.[21]

In some cases, fecal incontinence is a result of surgical complications, e.g., neorectum misplacement. In the majority of children, fecal incontinence is a result of an intrinsic anatomic defect. In men, the greatest incidence of incontinence is present in cases with rectal fistulas with communication to the bladder neck, while in women, high confluence cloacal malformation is most commonly associated with poor continence outcomes.[4] In congruence with Pelizzo et al.'s study, patients who underwent definitive surgical management within the first three months of life had higher rates of fecal continence.[6] Hakalmaz et al., however, mentioned inconsistent long-term rates of incontinence between 16% and 76% in ARM patients.[20] Postoperative continence was achieved in 70.8% of the patients in the present study.[15]

In a study by Hakalmaz et al., 75% of the patients' ARM were able to have voluntary bowel movements (VBM), and almost half became continent without adjunctive treatment.[20] The six low-birth-weight patients in this study did not have success with VBM. Malnutrition, a phenomenon common in low birth weight, is recognized to be a possible factor for compromised neuromuscular function. Voluntary bowel movements rely upon the integrity and coordinated function of pelvic floor musculature, rectum, and anal sphincter, all of which depend on sufficient innervation. Malnutrition may compromise the latter and functional maturation and potentially aggravate the severity of defective dysfunction in low-birth-weight infants with ARM compared to their normal-birth-weight peers. Previous research has also established a positive correlation between increased birth weight and improved survival rates among ARM patients. Defective voluntary control over the bowel may also occur secondary to anatomical defects or as a complication of reconstructive surgical procedures that affect the pelvic floor, rectum, or sphincteric mechanisms.[2]

The frequency of constipation following definitive repair of ARM has ranged extensively in the literature from less than 22% to as high as 86%.[20] The frequency of constipation following surgery was relatively low in the present study, occurring in only 7.6% of patients. The pathophysiology of constipation in ARM patients is not entirely clear. One of the proposed mechanisms is interference with rectal sensory innervation from the extensive anorectal mobilization in surgery, which could cause reduced rectal sensation. Hypomotility of the rectosigmoid area has also been implicated. Constipation also seems to be more common in operations that involve preservation of the internal anal sphincter, perhaps because of changed anorectal reflexes and motility.[4]

Chronic dilation of the rectal pouch may disrupt effective defecation by impairing peristaltic function, thus causing constipation.[2,7,9] It is one of the most prevalent and aggravating postoperative complications in patients with ARM who undergo definitive surgical repair.[1,8,14] Although constipation would be theoretically more prevalent in low-type anomalies, due to preservation of the internal anal sphincter and increased rectal reservoir function, it is often seen in both low and high ARM subtypes. In the study by Narad et al., a higher prevalence of constipation was reported in recto-bulbar and recto-prostatic fistulas, and to a lesser extent in recto-vesical fistulas. Effective management of constipation is necessary since it can otherwise lead to overflow fecal incontinence and gradual rectal or sigmoid dilation (megarectum or megasigmoid), significantly complicating subsequent treatment.[15]

In our research, approximately 61.5% of the patients achieved voluntary bowel movements with satisfactory sphincter control; however, a subgroup of patients continued to experience soiling episodes. Soiling developed in 11.5% of patients, less than in some past series.[1] Soiling is usually attributed to overflow incontinence, possibly due to sphincteric dysfunction or chronic constipation.[9] The height of the rectal blind pouch or fistula is usually inversely correlated with functional outcomes, with higher lesions correlated with poorer continence.[17] Lower-type anorectal malformations are usually preceded by a more favorable functional outcome than higher lesions.[2,22] It needs to be remembered that four patients (15.4%) of this series underwent minimal anoplasty, whose introduction of potential bias must be taken into account when assessing postoperative continence outcomes.

Despite advances in the surgical techniques for anorectal malformations, a subset of patients continues to experience severe postoperative complications, including overflow incontinence secondary to constipation and bowel motility abnormalities.[8] It should be kept in mind that the present study was not able to consider a variety of other possible factors that could influence functional outcomes following definitive repair. These include the presence of associated

anomalies, sacral and spinal morphology, value of sacral ratio, variability in surgical methods, and intra- or postoperative complications. Therefore, QOL evaluation must be included in the comprehensive follow-up of ARM patients, as reported in some of the earlier studies. Anatomic considerations, such as the nature of malformation and repair, are merely one group of factors, however, that influence fecal continence and QOL; sensory function and motility are also significant.[6]

CONCLUSIONS

Anorectal malformations (ARM) in our society are frequent. According to the Krickenbeck continence score used at our hospital, the functional outcomes for patients with ARM are generally positive. More than fifty percent of the children achieve bowel management (VBM), and relatively a few experience issues with soiling or constipation. Furthermore, the frequency of achieving VBM may be linked to factors such as gender and birth weight, but not to the ARM types. In contrast, constipation and soiling don't seem to be linked to gender, birth weight, or the type of malformation.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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