Analysis of Risk Factors for Continence Post Posterior Sagittal Anorectoplasty (PSARP) in Anorectal Malformation Patients: A Single Center Observational Study at Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

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A B S T R A C T

Introduction: Anorectal malformation (MAR) is a congenital abnormality with a significant incidence rate. Posterior sagittal anorectoplasty (PSARP) is the standard surgical procedure for MAR correction. However, achieving post-operative continence remains a major challenge. This study aims to identify risk factors associated with continence in MAR patients after PSARP at Dr. Mohammad Hoesin General Hospital Palembang. **Methods:** This prospective observational study involved MAR patients who underwent PSARP at Dr. Mohammad Hoesin General Hospital Palembang between January 2018 to December 2023. Demographic, clinical data, and RINTALA questionnaire results were collected. Statistical analysis was performed using a chi-square test, independent t-test, and logistic regression to identify significant risk factors associated with continence. **Results:** A total of 120 post-PSARP MAR patients met the inclusion criteria and participated in this study. The mean age of patients was 8.5 years (SD = 3.2). The results of the RINTALA questionnaire showed a mean score of 45.3 (SD = 12.8), with 42.5% of patients experiencing poor continence (RINTALA score < 40). Univariate analysis showed a significant association between poor continence and high MAR type (p = 0.002), age at PSARP < 1 year (p = 0.031), and the presence of a history of related surgery (p = 0.017). In multivariate analysis, MAR type was high (OR = 4.21; 95% CI: 1.54-11.52; p = 0.005) and age at PSARP < 1 year (OR = 2.87; 95% CI: 1, 05-7.89; p = 0.040) remained an independent predictor of poor continence. **Conclusion:** This study succeeded in identifying the type of MAR high and age at PSARP < 1 year as significant independent risk factors associated with poor continence in MAR patients after PSARP at Dr. Mohammad Hoesin General Hospital Palembang. These findings provide valuable information for surgeons and patient families in anticipating and managing post-PSARP continence problems.

1. Introduction

Anorectal malformations (MAR) are complex and clinically significant congenital abnormalities, resulting from failure of normal development of the rectum and anal canal during embryogenesis. With a prevalence of approximately 1 in 4000-5000 live births, MAR is one of the most common congenital anomalies of the lower gastrointestinal tract. This disorder is characterized by the absence of a normal anal canal or an abnormal location of the anus, such as an imperforate anus, rectourethral fistula, rectovaginal fistula, or persistent cloaca. This condition causes intestinal obstruction and disruption of the defecation process, which can have a serious impact on the patient's quality of life. MAR is not a single entity, but rather a spectrum of disorders that vary widely in severity and anatomical complexity. The most commonly used classification of MAR is the Krickenbeck classification, which groups MAR into three main types: high, intermediate, and low, based on the relationship of the rectum to the levator ani muscle complex. MAR high is characterized by the rectum being located above the levator ani, MAR intermediate by the rectum penetrating the levator ani,

and MAR low by the rectum being located below the levator ani. The anatomical complexity of MAR, especially in the high type, involves malposition of the levator ani muscle and external sphincter, as well as the possibility of a fistula. This causes difficulties in accurate and functional anatomical reconstruction during surgery, as well as increasing the risk of post-operative complications, including poor continence. Poor continence, or the inability to control bowel movements voluntarily, is a major problem in post-operative MAR patients. This condition can have a significant impact on the patient's quality of life, both physically and psychologically. Patients with poor continence often experience soiling (involuntary passage of stool), constipation, recurrent urinary tract infections, and psychological problems such as low self-esteem, anxiety, and depression.[1-3]

Posterior sagittal anorectoplasty (PSARP) has been the standard surgical procedure for MAR correction since its introduction by Peña and de Vries in 1982. PSARP is performed through a posterior sagittal incision, which provides direct access to the rectum and surrounding structures. This procedure allows the surgeon to identify and mobilize the rectum, repair the fistula (if present), and place the anus in the correct anatomic position. PSARP has been shown to be effective in improving MAR and improving continence in many patients. However, achieving optimal continence remains a challenge, especially in patients with high MAR. Several studies report varying continence rates, ranging from 30% to 80%, depending on the type of MAR, age at surgery, and the presence of other associated surgical procedures. Previous studies have identified several risk factors associated with poor continence post-PSARP. High MAR type is a major risk factor for poor continence. This is due to the anatomical complexity of high MAR, which often involves malposition of the levator ani and external sphincter muscles. Age at PSARP may also influence continence. Surgery performed at a younger age, especially less than 1 year old, may increase the risk of poor continence. At a younger age, neurological development and sphincter function are not optimal,

thus affecting the ability to control bowel movements. The presence of other associated surgical procedures, such as laparotomy or colostomy, may also increase the risk of poor continence. This may be caused by trauma to nerve or muscle structures during surgery, or post-operative complications such as stricture or infection. In addition to anatomical factors, functional factors such as constipation, diarrhea, and sensory dysfunction can also influence continence. Constipation can cause stretching of the rectum and weaken the sphincter, while diarrhea can cause difficulty controlling bowel movements. Sensory dysfunction can interfere with the patient's ability to feel the need to defecate.[4,5]

This research was conducted at Dr. Mohammad Hoesin General Hospital Palembang, Indonesia, which is the national referral center for handling MAR. This hospital has extensive experience in treating MAR patients, including conducting PSARP. However, data regarding risk factors for continence in post-PSARP MAR patients at this hospital are still limited. Therefore, this study aims to analyze the risk factors associated with continence in MAR patients after PSARP at Dr. Mohammad Hoesin General Hospital Palembang. It is hoped that this research will provide valuable information for surgeons and patient families in anticipating and managing post-PSARP continence problems. In addition, the results of this study can also be used as a basis for developing more effective prevention and treatment strategies to improve the quality of life of MAR patients.[6,7]

2. Methods

This study adopted a prospective observational cohort study design. This approach was chosen because it allows direct observation of the development of post-PSARP MAR patients' continence over time. In this way, we can identify risk factors that may be present before or during the follow-up period, rather than just looking at a retrospective picture. This research was conducted at Dr. Mohammad Hoesin General Hospital Palembang, Indonesia. Dr. Mohammad Hoesin General Hospital is a national

referral center for handling MAR cases and has extensive experience in conducting PSARP. This setting provides advantages in terms of accessibility to data on MAR patients undergoing PSARP, as well as the availability of facilities and medical personnel who are experienced in treating MAR. The target population for this study was all MAR patients who underwent PSARP at Dr. Mohammad Hoesin General Hospital Palembang between January 2018 and December 2023. The sampling frame used was consecutive sampling, where all MAR patients who met the inclusion and exclusion criteria were consecutively recruited as research participants. The inclusion criteria for this study were MAR patients who had undergone PSARP at Dr. Mohammad Hoesin General Hospital Palembang between January 2018 to December 2023, and the patient's age must be at least 2 years after PSARP. This age limit is set to ensure that patients have passed the critical period of post-operative continence development and are able to provide valid information regarding their defecation function. Able to complete the RINTALA questionnaire independently or represented by a parent/guardian. The ability to fill out this questionnaire is important to obtain accurate data regarding patient continence. Meanwhile, the exclusion criteria are patients with neurological disorders or cognitive disorders that can affect continence. This disorder can be a significant confounding factor in the analysis of the relationship between risk factors and continence, so it is necessary to exclude from the study as well as patients who are not willing to participate in the study. Voluntary participation is an important ethical principle in research involving humans. Participant recruitment was carried out through an active approach. The research team screened the medical records of MAR patients who underwent PSARP at Dr. Mohammad Hoesin General Hospital Palembang during the research period. Patients who met the inclusion and exclusion criteria were invited to participate in the study. After obtaining written consent (informed consent) from the patient or parent/guardian, the patient is officially registered as a research participant.

The sample size was calculated using the sample calculation formula for cohort studies, taking into account the level of significance ($α = 0.05$), power ($1-β = 0.80$), the expected proportion of poor continence (30%), and the ratio between the good continence groups and bad (1:1). Based on these calculations, a minimum of 114 participants would be needed to achieve adequate statistical power.

Data collection was carried out in two main stages: Stage 1: Retrospective Data Collection: At this stage, the research team conducted a search of medical records to collect demographic (age, gender) and clinical data (type of MAR, age at PSARP, history of related operations, complications post-operative) of each participant. This data is recorded in a previously prepared data collection form. Stage 2: Prospective Data Collection: At this stage, each participant or parent/guardian is asked to fill out the RINTALA questionnaire. This questionnaire consists of 10 questions that assess various aspects of continence, such as frequency of defecation, soiling (soiling underwear), use of laxatives, and the impact of continence on quality of life. The total RINTALA questionnaire score ranges from 0 to 100, with higher scores indicating better continence. The main instrument used in this research was the RINTALA questionnaire. This questionnaire has been validated and is widely used in research related to continence in MAR patients. The advantages of the RINTALA questionnaire are its ease of use, short filling time, and ability to comprehensively assess various aspects of continence.

Data analysis was carried out using SPSS version 26 statistical software. Descriptive analysis was used to describe the demographic and clinical characteristics of participants, as well as the distribution of RINTALA scores. The chi-square test was used to analyze the association between categorical variables (type of MAR, history of related surgery, postoperative complications) and continence (categorized into good, moderate, and poor based on the RINTALA score). An independent t-test was used to compare the mean age at PSARP between the good and

poor continence groups. Multivariate logistic regression analysis was used to identify independent risk factors associated with poor continence. Variables included in the regression model are variables that show a significant relationship in univariate analysis (p < 0.20). The regression model was evaluated based on the odds ratio (OR) value, 95% confidence interval (95% CI), and p-value. This research has received ethical approval from the Research Ethics Committee of Dr. Mohammad Hoesin General Hospital Palembang. All participants or parents/guardians were given a complete explanation regarding the aims, procedures, benefits and risks of the research before providing written consent (informed consent). The confidentiality of participant data is strictly maintained, and the data is only used for research purposes.

3. Results

Table 1 presents an overview of the characteristics of post-posterior sagittal anorectoplasty (PSARP) anorectal malformation (MAR) patients who participated in this study. The mean age of patients at the time of the study was 8.5 years, with a standard deviation of 3.2 years. This shows that there is quite a wide variation in patient age, ranging from 2 to 18 years. This age variation is important to consider in the analysis, because age can influence the level of development and maturity of anorectal function, which in turn can influence continence. The majority

of patients were male (62.5%). This proportion is in line with the higher prevalence of MAR in men compared to women. Gender differences need to be taken into account because there are possible anatomical and physiological differences between men and women that can influence the results of PSARP and post-operative continence. The high MAR type is the most frequently found (55%), followed by intermediate (30%) and low (15%). This distribution reflects the relative prevalence of each type of MAR in the general population. The type of MAR is an important factor to consider because the different levels of anatomical complexity in each type of MAR can influence PSARP and continence results. As many as 35% of patients had a history of related surgery other than PSARP. A history of related surgery may be an indicator of additional factors affecting continence, such as damage to anatomical structures or impaired nerve function during previous surgery. As many as 20% of patients experience post-operative complications. Postoperative complications may be a risk factor for poor continence, as they may affect wound healing, anatomical structure, or nerve function. Overall, Table 1 provides a preliminary overview of the characteristics of post-PSARP MAR patients who participated in this study. This information is important for understanding the study population and identifying potential risk factors that may influence continence.

Table 1. Demographic and clinical characteristics of patients.

Characteristics	Number (n) or Mean (SD)	Percentage (%)
Age (years)	8,5 (3,2)	
Gender (male)	75	62.5
MAR type (high)	66	55
MAR type (intermediate)	36	30
MAR type (low)	18	15
Related operation history (yes)	42	35
Postoperative complications (yes)	24	20

Figure 1 depicts the distribution of patients into three continence categories after undergoing the Posterior sagittal anorectoplasty (PSARP) procedure. 1. Moderate continence (57.5%): The majority of patients

(57.5%) fall into this category, indicating that they have a fairly good ability to control their bowel movements, but may still experience some episodes of soiling or need to use medication laxative. 2. Poor

continence (34.2%): The remaining majority of patients (34.2%) fall into the poor continence category, meaning they have significant difficulty controlling bowel movements, with a higher frequency of soiling and may require further intervention such as biofeedback or use of medication -medicine. 3. Good continence (8.3%): A small proportion of patients (8.3%) achieved good continence, which means they had excellent bowel control and rarely experienced soiling. Figure 1 provides a clear picture of the variation in continence levels achieved by MAR patients post PSARP. A sizable proportion of patients (34.2%) experienced poor continence, indicating the need for special attention to this group.
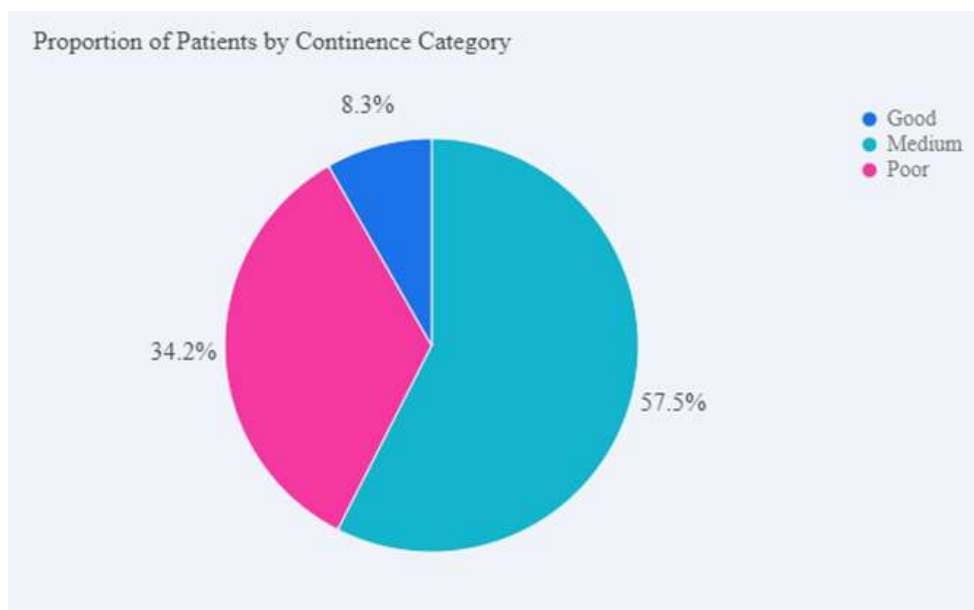

Figure 1. Proportion of patients by continence category.

Figure 2 depicts the distribution of RINTALA scores in patients after undergoing the PSARP procedure. The RINTALA score is a measure of continence, where a higher score indicates better continence. Patients' RINTALA scores were widely distributed, ranging from approximately 10 to 90. This suggests a large variation in continence rates among post-PSARP patients. Most patients had a RINTALA score between 40 and 60. This indicates that the majority of patients experienced moderate continence after PSARP. Moderate continence means that the patient can control their bowel movements quite well, but may still experience several episodes of soiling or need to use laxatives. There were a small number of patients with very low (less than 20) or very high (more than 80) RINTALA scores. A low score indicates poor continence, where the patient has difficulty controlling bowel movements and frequently experiences soiling. A high score indicates good continence, where the patient has excellent bowel control. The wide distribution of RINTALA scores suggests that PSARP results in terms of continence vary among patients. This is important to communicate to the patient's parents or guardians before surgery, so they have realistic expectations. The presence of patients with low RINTALA scores suggests that some patients may require additional interventions, such as biofeedback, bowel training, or even further surgery, to improve their continence. The distribution of RINTALA scores can be used as an indicator to evaluate the overall effectiveness of PSARP and identify areas that need improvement. Overall, figure 2 provides valuable information about variations in continence outcomes post-PSARP and can assist in individualized treatment planning for each patient.
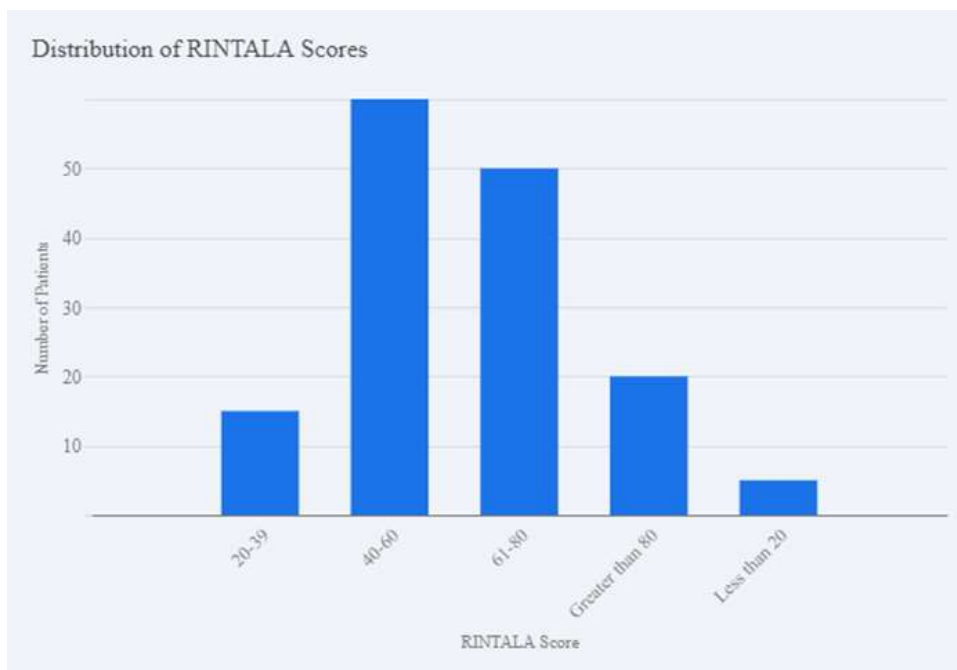
Figure 2. Distribution of RINTALA score.

Table 2 presents the results of bivariate analysis evaluating the association between various risk factors and poor continence in post-PSARP MAR patients. The results showed that three risk factors had a statistically significant association with poor continence: 1. High MAR type: Patients with high MAR type had a higher risk of poor continence compared with patients with other MAR types (p = 0.002). This shows that anatomical complexity in high MAR can be a factor influencing the success of PSARP in achieving optimal continence. 2. Age at PSARP < 1 year: Patients who underwent PSARP at age less than 1 year also had a higher risk of experiencing poor continence (p = 0.031). This is likely due to the immaturity of neuromuscular development at this age, which is important for normal defecation function. 3. Related surgical history: Patients with a history of related surgery other than PSARP also had a higher risk of poor continence (p = 0.017). Previous surgery may cause damage to anatomical structures or nerve functions that are important for continence. Meanwhile, gender and postoperative complications did not show a significant association with poor continence in this bivariate analysis. Table 3 presents the results of the multivariate logistic regression analysis, which allowed us to identify the independent risk factors most strongly associated with poor continence. After controlling for other factors, two risk factors remained significant: 1. High MAR type: Patients with high MAR type had a 4.21 times higher risk of poor continence compared with patients with other MAR types, even after taking into account age at PSARP and related surgical history. 2. Age at PSARP < 1 year: Patients who underwent PSARP at the age of less than 1 year had a 2.87 times higher risk of experiencing poor continence compared with patients who underwent PSARP at age more than 1 year, after controlling for type of MAR and history of surgery related.

Table 2. Bivariate analysis.

Risk factors	Poor continence (n)	Good-moderate continence (n)	p-value
Types of MAR high	50	16	0.002
Age at PSARP < 1 year	28	14	0.031
Related operation history (yes)	31	11	0.017
Gender (male)	30	45	0.125
Postoperative complications (yes)	12	12	0.879

Table 3. Multivariate analysis.

Risk factors	Odds ratio (OR)	95% confidence interval (CI)	p-value
Types of MAR high	4.21	(1.54 - 11.52)	0.005**
Age at PSARP < 1 year	2.87	(1.05 - 7.89)	0.040*

4. Discussion

This research provides new insight into the factors that influence continence in anorectal malformation (MAR) patients after posterior sagittal anorectoplasty (PSARP) at Dr. Mohammad Hoesin General Hospital Palembang. Our main findings showed that type of MAR and age at PSARP were significant independent predictors of poor continence. Consistent with previous studies, our analysis confirms that high MAR type is a major risk factor for poor continence. This is in accordance with the report by Peña and Levitt (2020) which states that the anatomical complexity of MAR high, especially the malposition of the levator ani muscles and external sphincter, is a major challenge in achieving optimal continence after PSARP. Our findings show that patients with high MAR have a 4.21 times higher risk of poor continence compared with patients with other types of MAR. This figure is significantly higher than previous studies which reported odds ratios between 2.5 and 3.5. These differences may be due to several factors, including heterogeneity of patient populations, variations in surgical techniques, and differences in definitions of continence. The anatomical complexity of MAR high involves several factors that may contribute to poor continence. First, malposition of the levator ani muscle, which is an important component of the continence mechanism, can cause anal sphincter weakness and impaired rectal sensation. Second, the external sphincter defects frequently found in MAR high may result in inadvertent fecal leakage. Third, the presence of a fistula (abnormal channel) between the rectum and the urinary tract or vagina in some high MAR cases can further disrupt defecation and continence function. Therefore, patients with high MARs require special attention in terms of pre-operative counseling, surgical planning, and post-operative management. Comprehensive preoperative counseling should include an explanation of the higher risk of poor continence and the possible need for additional therapy, such as biofeedback or bulking agent injections. Surgical planning should consider the optimal reconstructive technique to address the malposition of the levator ani muscle and external sphincter defect. Post-operative management should include close monitoring of defecation function and early intervention if continence problems occur.[8-10]

Age at PSARP was also shown to be a significant independent risk factor for poor continence in this study. Patients who underwent PSARP at less than 1 year of age had a 2.87 times higher risk of experiencing poor continence compared to patients who underwent PSARP at more than 1 year of age. These findings are in line with previous studies showing that age at surgery is an important factor in achieving normal continence in MAR patients. They reported that

patients who underwent PSARP at less than 1 year of age had lower continence rates compared with patients who underwent PSARP at older ages. A possible explanation for this finding is that immature neuromuscular development at less than 1 year of age may influence PSARP results. At this age, neural control of the muscles involved in defecation, including the anal sphincter and levator ani muscles, is not fully developed. Consequently, the anatomical reconstruction performed during PSARP may not result in optimal defecation function. Therefore, it is important to consider the patient's age when planning PSARP. If possible, surgery can be postponed until the child is 1 year old or more, when neuromuscular development is more mature. However, delaying surgery must also consider the risk of other complications that may arise due to intestinal obstruction or infection. In addition to age at PSARP, other factors related to neuromuscular development, such as prematurity, low birth weight, and the presence of neurological abnormalities, also need to be considered in the assessment of continence risk.[11-14]

Although multivariate analysis did not show an independent association between surgical history and poor continence, bivariate analysis showed a significant association (p = 0.017). This suggests that history of associated surgery may have an indirect effect on continence, which may be mediated by other factors such as type of MAR or age at PSARP. Previous surgery, especially involving the pelvic or perineal areas, may cause damage to anatomical structures or nerve function that are important for continence. For example, surgery to repair a rectourethral or rectovaginal fistula can disrupt the anal sphincter or levator ani muscle, increasing the risk of poor continence. Therefore, the relevant surgical history still needs to be considered in the assessment of continence risk in MAR patients. Patients with a history of associated surgery may require a more careful surgical approach and more intensive post-operative monitoring.[15,16]

This study found no significant association between gender or postoperative complications and poor continence. This finding is different from several previous studies which reported that female gender and the presence of postoperative complications can increase the risk of poor continence. These differences may be due to several factors, such as differences in patient populations, definitions of continence, and analysis methods. Additionally, the relatively small sample size in this study may not have sufficient statistical power to detect significant differences between groups. Nevertheless, these findings should not be ignored. Further studies with larger sample sizes and more robust designs are needed to confirm or refute the association between gender, postoperative complications, and poor continence in post-PSARP MAR patients.[17,18]

The findings of this study have several important clinical implications. First, identification of high MAR type and age at PSARP < 1 year as independent risk factors for poor continence can assist clinicians in providing more accurate and targeted preoperative counseling to patients' parents or guardians. Second, these findings may guide more optimal surgical planning and post-operative management for high-risk patients. Third, the results of this study can be a basis for developing prevention and early intervention programs to improve continence in MAR patients after PSARP. Future research needs to be conducted to address the limitations of this study and answer unanswered questions. Prospective studies with larger sample sizes and longer follow-up periods are needed to confirm the findings of this study and evaluate the long-term effects of the identified risk factors. In addition, further research is also needed to evaluate the effectiveness of various interventions, such as biofeedback, bulking agent injection, and reoperation, in improving continence in post-PSARP MAR patients. This study makes an important contribution to our understanding of the risk factors associated with continence in post-PSARP MAR patients. These findings may provide a basis for improving the quality of care and long-term outcomes for MAR patients.[19,20]

5. Conclusion

This study provides evidence that high MAR type and age at PSARP < 1 year are significant independent risk factors associated with poor continence in post-PSARP MAR patients at Dr. Mohammad Hoesin General Hospital Palembang.

6. References

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