



Original article

Functional Outcomes Following Posterior Sagittal Anorectoplasty According to Pena for High and Intermediate Anorectal Malformations: A Cross-Sectional Study at Charles De Gaulle University Hospital in Ouagadougou

Résultats fonctionnels après anorectoplastie sagittale postérieure selon Pena pour malformations anorectales hautes et intermédiaires : étude transversale au CHUP Charles De Gaulle de Ouagadougou

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Résumé

Introduction : La malformation anorectale (MAR) est une anomalie congénitale rare dont l'anorectoplastie sagittale postérieure (PSARP) selon Pena constitue la technique de référence. La principale complication post-opératoire reste l'incontinence anale. Cette étude a pour objectif principal d'évaluer les résultats fonctionnels après anorectoplastie sagittale postérieure (PSARP) selon Pena au service de chirurgie pédiatrique du CHU Charles de Gaulle de Ouagadougou.

Méthodologie : il s'agit d'une étude transversale descriptive réalisée dans le service de chirurgie pédiatrique du CHU Charles de Gaulle de Janvier à avril 2022 concernant tous les patients avec une MAR haute ou intermédiaire ayant bénéficié des 3 temps chirurgicaux entre Janvier 2010 à Décembre 2017. L'évaluation de la continence a été faite selon les critères de Krickenbeck sur un recul moyen de 8 ans. Résultats : nous avons colligé 32 patients dont 24

(75 %) avec MAR haute et 8 (25 %) avec MAR intermédiaire. Une émission volontaire de selle a été retrouvée chez 26 patients (81,25 %), une souillure chez 18 patients (56,25 %) dont 11 présentaient une souillure de grade 1, 4 une souillure de grade 2 et 3 seulement avec une souillure de grade 3. Il a été noté aussi une constipation chez un patient (3,13%).

Conclusion : la PSARP selon Pena octroie des résultats fonctionnels acceptables pour la prise en charge des MAR hautes et intermédiaires dans notre contexte.

Mots-clés : MAR, anorectoplastie, incontinence.

Abstract

Introduction: Anorectal malformation (ARM) is a rare congenital anomaly for which posterior sagittal anorectoplasty (PSARP) according to Pena is the standard procedure. The main postoperative complication remains anal incontinence. The primary objective of this study is to evaluate functional outcomes following posterior sagittal anorectoplasty

(PSARP) according to Pena in the pediatric surgery department at Charles de Gaulle University Hospital in Ouagadougou.

Patients and Methods: This is a descriptive cross-sectional study conducted in the pediatric surgery department of the Charles de Gaulle University Hospital from January to April 2022, involving all patients with high or intermediate ARM who underwent the three-stage surgical procedure between January 2010 and December 2017. Continence was assessed according to the Krickenbeck criteria over an average follow-up period of 8 years.

Results: We included 32 patients, of whom 24 (75%) had high-grade ARM and 8 (25%) had intermediate-grade ARM. Voluntary stool passage was observed in 26 patients (81.25%), fecal soiling in 18 patients (56.25%), including 11 with grade 1 soiling, 4 with grade 2 and 3 with grade 3 soiling. Constipation was also noted in one patient (3.13%).

Conclusion: Pena's PSARP provides acceptable functional outcomes for the management of high and intermediate ARM in our setting.

Keywords: ARM, anorectoplasty, incontinence.

Introduction

Anorectal malformations (ARM) or anorectal dysgenesis represent an anomaly in the process of establishment of the anorectal tract. These are malformations that occur early during intrauterine life and can therefore be part of a varied set of malformations [1,2].

Although rare (1/5000 births), ARM is the most frequent malformation affecting the perineal region [3,4].

The diagnosis of ARM in its isolated form is exceptional during pregnancy [2]. At birth, every newborn should undergo a perineal examination; this examination must be even more meticulous in girls to avoid missing a fistula [2].

The first description of an anorectal malformation repair dates back to the second century AD by

Soranus. Since then, several works have been carried out developing various repair techniques for ARM. In 1953, the Stephens Douglas team described sacroperineal anorectoplasty and highlighted the importance of the levator ani muscles. Today, the gold standard for ARM treatment is the Posterior Sagittal Anorectoplasty (PSARP) introduced by Professor Alberto Pena in 1982 [2].

All these techniques aim to address the problem of continence. The ultimate goal being to have patients with a socially acceptable life after pull-through. It has therefore become necessary for pediatric surgeons to evaluate their results in terms of continence, hence the development of several evaluation classifications. The last study on functional evaluation of ARM in Burkina Faso dates back to 2002 by Lanou H, who obtained 62.5% good results after a follow-up of 3.6 years [4]. To our knowledge, no study in Burkina Faso has evaluated functional outcomes after PSARP according to PENA.

This study was conducted with the aim of updating data on functional evaluation of ARM after PSARP according to Pena in the pediatric surgery department of the CHUP Charles de Gaulle of Ouagadougou.

Methodology

This is a descriptive cross-sectional study conducted at the CHUP Charles de Gaulle of Ouagadougou during the month of January to April 2022. The study included all patients with high or intermediate ARM who underwent the three-stage surgical procedure during the period from January 2010 to December 2017. The three surgical stages consisted of a diverting colostomy, anorectal pull-through by posterior sagittal anorectoplasty of Pena, and closure of the colostomy, respecting the series of anal dilatations until the desired caliber was reached.

Continence evaluation was performed at age ≥ 4 years. It was carried out in the consultation room; patients accompanied by parents responded to a standardized questionnaire according to the evaluation criteria for ARM repair adopted at the Krickenbeck

conference in 2005 [5]. The table below describes its evaluation criteria.

Table I: Criteria for evaluating anorectal continence according to the Krickenbeck classification

Criterion	Response	Grade	Description
1. Voluntary stool passage	Yes/No	-	Ability to verbalize urge to defecate, ability to hold stool, scheduled bowel movement possible
2. Soiling	Yes/No	Grade 1 Grade 2 Grade 3	Occasional (1-2x/week) Daily, without social impact Constant, social problem
3. Constipation	Yes/No	Grade 1 Grade 2 Grade 3	Manageable by dietary regimen Requires laxatives Resistant to diet and laxatives

Note: the Krickenbeck score does not produce a global score. The evaluation covers these 3 independent criteria. International conference, 2005.

Nevertheless, it is unanimously recognized that patients who obtained a good result have generally been considered socially continent, which implies that intestinal function defects do not lead to significant social disadvantage [6].

Excluded from our series were patients under 4 years of age from the definitive closure of their stoma and those whose parents and child were not available to answer the pre-established questionnaire.

Data were collected based on individual forms including the various parameters: age, sex, presence or absence of a fistula, WAGENSTEIN and RICE invertogram, spine radiograph (frontal and lateral), type of ARM, presence or absence of associated malformations, type of surgical treatment, duration of management, Pena dilatation sessions starting from the 10th postoperative day, continence evaluation according to the Krickenbeck classification.

Total continence is defined by a patient presenting voluntary stool passage without soiling or constipation, and we expect by complete surgical treatment a patient who has benefited from the 3 operative stages. The collected elements were encoded and analyzed using Epi Info 7.2.4.0 and Microsoft Office Excel 2016 software.

Anonymity and confidentiality of information collected as part of this study were preserved. Informed oral consent from parents for each participant was also obtained.

Results

Thirty-two patients were included in the study over a period of 4 months. Twenty-one patients (65.63%) were male and 11 patients (34.38%) were female, giving a sex ratio of 1.91. According to the Stephens classification, high ARM were found in 24 patients (75%). A fistula was present in 21 patients (65.63%). The table II shows the type of fistula found.

The invertogram was performed in 11 patients (34.38%) and all these patients presented with high ARM.

Twenty-one patients underwent spine radiography as part of the malformation assessment. No patient presented with an associated malformation.

Thirty patients (93.75%) had their colostomy before the age of 1 month. The colostomy was performed at an average age of 8 days with extremes of 1 and 120 days and a standard deviation of 22 days.

The mean age at the time of posterior sagittal anorectoplasty was 16 months with extremes of 4 and 108 months and a standard deviation of 19 months.

The table III describes the distribution of patients according to their age at anorectoplasty.

The mean duration of complete surgical treatment corresponding to the mean age at restoration of digestive continuity was 25 months with extremes of 7 and 115 months and a standard deviation of 20 months.

The patients were evaluated with an average follow-up of 8 years with extremes of 2 and 11 years and a standard deviation of 2 years.

Table IV represents the evaluation of patients

according to the Krickenbeck criteria.

The mean age of patients at the time of evaluation was 10 years with extremes of 7 and 12 years and a standard deviation of 2 years.

The table V shows the number of patients according to their age in years.

The functional results evaluated according to the Krickenbeck criteria are represented in the table VI.

The functional results in relation to the age at colostomy are represented in the table below.

Voluntary stool passage was found in 18 patients who underwent restoration before 24 months, i.e., 90% of

cases.

Voluntary stool passage was found in 26 patients, i.e., 86.67% of cases.

The table below describes the evaluation of the Krickenbeck score according to the age at restoration of digestive continuity (RCD).

Age RCD/voluntary stool passage: $p=0.2$ Age RCD/soiling: $p=0.5$ Age RCD/constipation: $p=0.3$

The functional results in relation to the patient's age at the time of evaluation are represented in the table below. Table XI describes the association between patient age and the elements of the Krickenbeck score.

Table II: Frequency of patients according to the type of fistula (n=32)

Fistula type	Number (n)	Percentage (%)
Rectourethral	10	31.25
Rectovaginal	01	3.13
Rectovestibular	10	31.25
No fistula	11	34.38
Total	32	100

Table III: Frequency of patients according to the age of anorectoplasty (n=32)

Age at anorectoplasty	Frequency	Percentage (%)
[0-6 months[02	6.25
[6-12 months[12	37.5
>= 12 months	18	56.25
Total	32	100

Table IV: Evaluation of patients according to the Krickenbeck criteria (n=32)

	Number (n)	Percentage (%)
Voluntary stool passage - Yes	27	84.38
Voluntary stool passage - No	5	15.63
Soiling - Grade 1	11	34.38
Soiling - Grade 2	4	12.50
Soiling - Grade 3	3	9.37
No soiling	14	43.75
Constipation - Grade 1	1	3.13
Constipation - Grade 2	0	0
Constipation - Grade 3	0	0
No constipation	31	96.88
Total continence	12	37.50

Table V: Frequency of patients according to age in years (n=32)

Age	Frequency	Percentage (%)
[7-9[08	25
[9-11[14	43.75
>= 11	10	31.25
Total	32	100

Table VI: Krickenbeck score according to the type of ARM (n=32)

ARM type	N	Voluntary stool Yes	Voluntary stool No	Soiling G1	Soiling G2	Soiling G3	Constipation G1
High ARM	24	19 (79.17%)	5 (20.83%)	9 (60%)	3 (20%)	3 (20%)	1 (100%)
Intermediate	8	8 (100%)	0	2 (66.67%)	1 (33.33%)	0	0

The significance threshold (p<0.05) according to the type of ARM is distributed as follows:

ARM type/voluntary stool passage: p=0.2

ARM type/soiling: p=0.2

ARM type/constipation: p=0.7

Table VII: Krickenbeck score according to the type of fistula (n=32)

Fistula type	N	Voluntary stool Yes	Voluntary stool No	Soiling G1	Soiling G2	Soiling G3	Constipation G1
Rectourethral	10	8 (80%)	2 (20%)	3	1	0	0
Rectovaginal	1	1 (100%)	0	0	0	0	0
Rectovestibular	10	9 (90%)	0	1	0	3 (50%)	0
No fistula	11	9 (81.82%)	2 (18.18%)	5	1	0	1 (100%)

Fistula yes/no/Voluntary stool passage: p= 0.046

Fistula yes/no/Soiling: p = 0.437

Fistula yes/no/Constipation: p = 0.004

Table VIII: Krickenbeck score and age at colostomy (n=32)

Age colostomy	N	Voluntary stool Yes	Voluntary stool No	Soiling G1	Soiling G2	Soiling G3	Constipation G1
[0-1 month]	30	26 (86.67%)	4 (13.33%)	11	4	3	2
>1 month	2	1 (50%)	1 (50%)	0	0	1	0

Age at colostomy/Voluntary stool passage: p= 0.2

Age at colostomy/Soiling: p = 0.6

Age at colostomy/Constipation: p = 0.9

Table IX: Krickenbeck score according to the age of anorectoplasty (n=32)

Age PSARP	N	Voluntary stool Yes	Voluntary stool No	Soiling G1	Soiling G2	Soiling G3	Constipation G1
[0-6 mo[2	2 (100%)	0	0	1	0	0
[6-12 mo[12	9 (75%)	3 (25%)	3	1	1	0
>=12 mo	18	16 (88.89%)	2 (11.11%)	8	2	2	2

Age PSARP/voluntary stool passage: p=0.090

Age PSARP/soiling: p=0.258

Age PSARP/constipation: p=0.010

Table X: Krickenbeck score according to the age of RCD (n=32)

Age RCD	N	Voluntary stool Yes	Voluntary stool No	Soiling G1	Soiling G2	Soiling G3	Constipation G1
[0-24 mo[20	18 (90%)	2 (10%)	10	1	1	0
>=24 mo	12	9 (75%)	3 (25%)	1	3	2	1 (100%)

Age RCD/voluntary stool passage: p=0.2 Age RCD/soiling: p=0.5 Age RCD/constipation: p=0.3

Table XI: Association between patient age and Krickenbeck score (n=32)

Age (years)	N	Voluntary stool Yes	Voluntary stool No	Soiling G1	Soiling G2	Soiling G3	Constipation G1
[7-9[8	7 (87.50%)	1	2	0	0	0
[9-11[14	10 (71.43%)	4	6	1	0	0
>=11	10	10 (100%)	0	3	0	0	1 (100%)

Discussion

The descriptive cross-sectional nature of our study allowed us to obtain a lot of clinical and paraclinical information. Nevertheless, we had difficulties in data collection with a lost to follow-up rate of nearly 66.7% due to geographic inaccessibility and lack of data digitization. This explains the small size of our sample. Also, functional data were collected in outpatient consultations based on parental responses, which may constitute bias because they may over or underestimate problems due to modesty or ignorance of what is considered normal. The heterogeneous follow-up also constitutes a limitation due to the difference in sphincter neurologic maturation variable according to age. The retrospective nature of our study in its data collection part, the unavailability of certain

complementary examinations such as manometry did not allow us to use other scores such as that of Rintala and Kelly. The appreciation of these scores would have resulted in a validated global score. The study was conducted in a single center due to insufficient qualified personnel and the country had only one surgical management center for children at that time. Despite these difficulties, we achieved results that we were able to compare with those of other authors in the literature, which allowed us to conduct a discussion. The Krickenbeck criteria allowed us to find voluntary stool passage in 26 patients, i.e., 81.25% of cases, uncontrolled defecation in 5 patients, i.e., 15.62% of cases, soiling was found in 18 patients, i.e., 56.25% of cases, constipation was found in one patient, i.e., 3.1% of cases, and global total continence in 12 patients, i.e., 37.5% of cases.

Among the 05 incontinent patients, one had undergone surgical revision for anal stenosis and two had undergone revision of the dilation protocol; which could explain their poor result. We noticed that a patient with good voluntary stool passage was not exempt from soiling; however, grade 1 soiling was predominant with 11 cases, the case of constipation was managed with an appropriate diet.

Table XII distributes the percentage of patients according to the elements of the Krickenbeck score by authors.

Table XII: Percentage of patients according to the elements of the Krickenbeck score by authors

Authors	Voluntary stool passage	Soiling	Constipation
Bensebti (2019, Algeria)	82.9%	48.5%	14.3%
Makrufardi (2020, Indonesia)	53.5%	53.5%	11.6%
Medina (2020, Indonesia)	83.3%	9.3%	9.8%
Our study	84.38%	56.25%	3.1%

Our results are similar to those of Bensebti [7] in Algeria (2019) and Medina [8] in Indonesia (2020) for voluntary stool passage but differ from Makrufardi [9] and Medina [8] for soiling.

This difference could be explained by the fact that to reduce the frequency of soiling, it is necessary to have good hygiene adapted to the child and good parent-child collaboration. Meeting these conditions is often difficult in developing countries due to the inaccessibility of care centers and parents who are not always available, making postoperative follow-up less regular.

Table XIII presents the frequencies of total continence according to authors.

Table XIII: Frequencies of total continence according to authors

Authors	Total continence	Percentage (%)
Bensebti (2019, Algeria)	17	48.6
Richard (1991, Netherlands)	3	8.1
Medina (2020, Indonesia)	14	19.4
Our study	12	37.5

Our result is similar to that of Bensebti [7] but different from that of Richard [10] and Medina [8]. Richard and Medina used a different evaluation score, that of RINTALA. This would explain this difference. Brisighelli et al [1] in 2017 in Italy had found that results could vary according to the evaluation score. They had compared several scores and found total continence in 10 patients with the Krickenberg score and in 3 patients with the Rintala score for high/intermediate ARM.

The anatomical form of the ARM is an essential element of prognosis. It is accepted that the higher the rectal pouch, the more the muscular and sphincteric structures, and potentially their innervation, are altered [2].

For high ARM, 19 patients, i.e., 79.17% of cases, acknowledged having voluntary stool passage, 7 patients, i.e., 29.1% of cases, had total continence.

For intermediate ARM, 8 patients acknowledged having voluntary stool passage, i.e., 100% of cases, with total continence in 5 patients, i.e., 62.5%.

Our result is in agreement with the literature data; intermediate ARM had better functional results than high

ARM, but we did not find a significant link between the type of ARM and voluntary stool passage ($p=0.2$). Pollakan et al [11] in 2017 in Thailand also found that intermediate ARM had better prognosis than high ARM. They obtained voluntary stool passage in 15 patients with high ARM, i.e., 51.7% of cases, and in 83 patients with intermediate ARM, i.e., 70.3% of cases, after a follow-up of 7 years. In this study, there was a significant link between the type of ARM and voluntary stool passage ($p=0.04$). Shahnam et al [12] in 2014 in Iran in their study compared the functional results of high and low ARM; they did not find a significant difference between the two forms using the RINTALA score, which could explain the difference in results with ours.

In our series, total continence was found in 9 patients with a fistula, i.e., 42.86% of cases, and 3 without a fistula, i.e., 27.27% of cases. Voluntary stool passage was found in 18 patients with a fistula, i.e., 85.71% of cases.

This result indicates that patients with a fistula have better functional results than those without. There was a significant link between voluntary stool passage and the presence of a fistula ($p=0.046$).

Shahnam [12] in 2014 in Iran had also found a higher score ($n=51, 13.94\pm 3.19$) therefore better functional results for ARM with fistula than for ARM without fistula ($n=9, 11.8\pm 3.3$; $P=0.03$); using the RINTALA score. In this study, there was no significant link between voluntary stool passage and the presence or absence of a fistula.

Makrufardi et al [9] in 2020 in Indonesia had found a different result from ours. 10 patients, i.e., 55.55% of cases without fistula, reported having voluntary stool passage, 13 patients, i.e., 52% of cases with fistula, reported having voluntary stool passage. There was no significant difference between the two groups and in their study there was also no statistically significant link between voluntary stool passage and the type of fistula ($p=0.26$). This difference is a simple statistical observation.

The level of the fistula conditions the functional prognosis. In boys, rectourethral (bulbar) fistulas

have good prognoses, as do rectovestibular fistulas in girls [2, 5].

In our study, 8 patients, i.e., 80% of cases with rectourethral fistula, reported having voluntary stool passage. Nine patients, i.e., 90% of cases with rectovestibular fistula, reported having voluntary stool passage. The patient with rectovaginal fistula had voluntary stool passage.

Our result is in agreement with the literature data; patients with rectourethral and rectovestibular fistula have good functional results, as does rectovaginal fistula.

Patel et al [13] in 2018 in India had found a similar result. Twelve patients, i.e., 80% of cases, had answered yes to voluntary stool passage for rectourethral fistulas and 26 patients, i.e., 83.8% of cases for rectovestibular fistulas. Makrufardi et al [9] in 2020 in Indonesia had also found a similar result for rectourethral fistulas; 8 patients, i.e., 72.72% of cases had answered yes to voluntary stool passage but obtained a different result from ours for rectovestibular fistulas; 1 patient, i.e., 25% of cases had answered yes to voluntary stool passage. This difference could be linked to the sample size since they had only evaluated 4 patients with rectovestibular fistula.

Early colostomy reduces the risk of rectal ectasia before surgical repair, which improves functional results [14].

Colostomy was performed in 30 patients in the first month of life. None of these patients presented with rectal ectasia; we found voluntary stool passage in 26 patients, i.e., 86.67% of cases.

Our result is satisfactory and in agreement with the literature data, although we did not find a significant link between the age at colostomy and the elements of the Krickbeck score ($p=0.2$).

Our result is similar to that of Kigo [14] in Kenya in 2002, who performed a colostomy in the first month of life in 104 patients, of whom 79 patients, i.e., 76% of cases, presented with voluntary stool passage.

Most authors agree on early radical treatment between 1 and 3 months to allow good staturo-ponderal development [15]. PENA [16] states that pull-through

between the 6th and 8th week of life is possible depending on the weight and general condition of the patient.

In our study, we performed pull-through in two children before the age of 6 months; both patients reported having total continence.

Between 6-12 months, 12 patients underwent pull-through and 9 cases, i.e., 75%, reported having good voluntary stool passage. Four patients had total continence.

After 12 months, 18 patients underwent pull-through and 16 cases, i.e., 88.8%, reported having voluntary stool passage. Six patients had total continence.

Patients operated earlier (before 6 months) obtained better functional results, but we did not find a significant link between the age at PSARP and voluntary stool passage ($p = 0.09$).

Our result is similar to that of Bensebti [7] in 2019 in Algeria, who obtained voluntary stool passage in 92.3% of cases in patients operated at less than 6 months versus 55.6% in patients operated after 6 months. Kigo [14] in 2002 in Kenya found in his study that voluntary defecation was 81.1% when radical treatment was done between 0 and 6 months and that this percentage decreased as surgery was performed later.

Rapid restoration of digestive continuity allows early adaptation to the evacuation function and neurologic maturation of this function with advancing age, which offers a better chance of good continence [7].

We found 20 patients who were restored before 2 years. Among them, 18 cases, i.e., 90%, reported having voluntary stool passage and 8 patients, i.e., 40% of cases, had total continence. Twelve were restored after 2 years, of whom 9 cases, i.e., 75%, reported having voluntary stool passage, 4 patients, i.e., 33.33% of cases, had total continence. Our results are in agreement with the literature; children who were restored before 2 years presented better functional results, but we did not find a significant link between the age at restoration and voluntary stool passage ($p = 0.2$).

Kyrklund [17] and Rintala [6] state that intestinal

function improves with age, while Pena asserts that intestinal function does not improve but patients become more aware and more interested in being socially continent [1].

In our series, the oldest patients (11 years or more) had better functional results. One hundred percent of cases had voluntary stool passage. This could be due either to adaptation or to restoration of intestinal functions.

Bensebti [7] had found a similar result; in his series total continence improved with age (36% at 3 years versus 56% at 4 years). Similarly, Kyrklund [17] in 2016 in Finland had found that continence improved with age (prevalence of soiling was 29% in patients aged 4-12 years; $p = 0.08$ versus 18% in those over 12 years).

Lanou [4] in 2002 had obtained 62.5% good results with a different score over an average follow-up of 3.6 years. We observe in our study a significant improvement with 81.25% voluntary stool passage and 37.5% total continence. This could be explained by the progress made in surgical management and long-term follow-up of these patients.

Conclusion

Anorectal malformation remains a real challenge for the surgeon. The use of PSARP according to Pena requires good knowledge of perineal anatomy in order to minimize complications and provide the patient with a socially acceptable life. This technique has allowed us to obtain acceptable functional results in our series.

The presence of a fistula, an intermediate ARM, and restoration of digestive continuity before two years seem to be associated with better results, although these associations did not reach statistical significance. The limitations of our study were its single-center nature with a small sample size.

However, in order to establish a formal relationship between PSARP and functional outcomes, it would be necessary to have a multicenter study with a larger sample to confirm these results.

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