

# Challenges, Constraints and Failures That are Related to the Posterior Sagittal Anorectoplasty Approach to Anorectal Malformations in a Low-Resource Context: An Experience from a Sudanese Tertiary Referral Centre

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## Abstract

**Background:** Anorectal malformations (ARMs) in the sub-Saharan Africa are a common cause of neonatal referral for intestinal obstruction, and the posterior sagittal anorectoplasty (PSARP) approach is rapidly spreading. The small number of paediatric surgeons and the low-resource context limit children's access to care and constrain the quality of results. A retrospective, observational study has been done on a consecutive series of ARM cases admitted to a Sudanese tertiary paediatric surgical centre within the framework of a partnership between Italian and Sudanese academic institutions addressed to review and upgrade the standard of care of major congenital anomalies. **Materials and Methods:** The authors collected 94 ARM cases in a 3 years' period. Conditions on referral, operative procedures, post-operative course and follow-up were recorded and examined. Their correlations with complications and outcome were analysed. **Results:** The male/female ratio was 47/47. Eighty patients presented with an untreated ARM; 66 had a divided stoma and 14 had already a PSARP procedure, followed by a poor outcome or sequelae. In 25% of the cases, colostomy required re-doing. In 57 cases, a staged PSARP (primary or re-do) was done. Surgical-site infections occurred in nine patients. Some patients were lost to follow-up after preliminary colostomy. Post-operative dilatation programme suffered from the lack of systematic follow-up, and colostomy closure was possible in 46% of the cases due to problems in travelling and accessing hospital care. Anal stenosis was frequently observed among unfollowed patients. **Conclusion:** Despite PSARP's widespread adoption in Africa, the risk of complications and failures is high. Primary management is often inappropriate, and a high rate of colostomy-related complications is observed. Poverty and lack of transportation reduce attendance to follow-up, hampering the final results. Investments in healthcare facilities and retention of trained health providers are needed to improve the standard of care.

**Keywords:** Africa, anorectal malformations, child, low-resource context, surgery

## INTRODUCTION

Anorectal malformations (ARMs) are the most common congenital intestinal defects among African neonates.<sup>[1]</sup> They represent a significant load on the paediatric surgical resources of the sub-Saharan Area.<sup>[2,3]</sup> A low number of trained surgeons concentrated in main urban centres, lack of transport facilities for the people living in rural areas and local healthcare

providers unable to deal with these patients properly, all contribute in making the survival uncertain and the quality of life miserable, of many children with ARMs.<sup>[4]</sup>

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The posterior sagittal anorectoplasty (PSARP) approach to ARMs<sup>[5]</sup> is diffused worldwide and can guarantee an acceptable outcome according to the severity of anatomical derangement. This approach is attractive for paediatric surgeons, and it is spreading over most of the African centres. Unfortunately, there are severe constraints to its application in a low-resource context, which can affect the quality of results. To upgrade the standard of care for selected paediatric surgical conditions, including ARMs, by close cooperation in teaching and training, a bilateral academic partnership programme has been established between the Pediatric Surgical Department of the University of Chieti-Pescara, Italy, and the Gezira National Centre of Pediatric Surgery (GNCPS)-Wad Medani, Sudan. The GNCPS is the only tertiary referral centre in this field outside the capital, Khartoum. It serves an area where about ten millions of people live, with about 58% of them in the paediatric age group. The programme is co-financed and supported by the Italian Agency for Development Cooperation (AICS), the GNCPS and the University of Gezira.

The present paper is the result of an observational study conducted on all patients with ARMs admitted at the GNCPS between October 2017 and February 2020. Italian and Sudanese partners reviewed the current practices and result in the management of ARMs by PSARP approach, identifying challenges and constraints and proposing solutions to improve surgical treatment and nursing care of patients to achieve a better outcome and quality of life.

## MATERIALS AND METHODS

We included 94 patients with different types of ARMs. Of which, eighty (Group A) had an untouched anomaly, and 66 had a diverting colostomy. Another 14 patients (Group B) were referred to after performing PSARP at the GNCPS, followed by sequelae or poor functional outcome. Two of them had a diverting colostomy. Variables recorded included gender, age, type of anomaly, associated defects and type of colostomy when present. Eligibility of patients in Group A to PSARP approach was evaluated considering the type of the anomaly and pre-operative conditions (associated defects, presence and quality of colostomy). PSARP re-doing versus alternative procedures were considered in Group B to ensure a better quality of life to patients. Variables influencing PSARP post-operative course were analysed using Fisher's exact test (GraphPad Prism 8.4.0 (San Diego, CA, USA)). The quality of follow-up was examined, and the causes of low parental compliance were also investigated. Bowel function after colostomy closure was reported.

## RESULTS

### Sex distribution and age

The male/female ratio was 39/41 among the eighty cases in Group A. The mean age at referral was 1 year and 9 months (median: 1, SD: 3.07). It ranged from 3 days of a male neonate to 23 years of a late referred Cloaca. Among

14 children of Group B, the mean age was 6 years and 4 months (range: 2 years–10 years, median: 4, SD: 2.92). The male/female ratio in this group was 8/6.

Table 1 reports the types of ARMs observed.

### Associated abnormalities

There were four patients with Down syndrome (incidence 4.25%). One had a small interventricular defect. A left, asymptomatic, obstructive megaureter was observed.

### Colostomy

A colostomy on the left sigmoid was already present in 35/39 males and 31/41 females in Group A. Among them, there was a 14-year-old male patient; the other 17 children (8 males and 9 females) had been waiting from 3 to 5 years for a PSARP procedure. About two-thirds of colostomies were done at the GNCPS and the others in a rural/district hospital. In nine cases (15%), stools came through the lower stoma due to intraoperative twisting of the sigmoid loop, and in seven of them, this caused a short distal loop. A prolapse of the distal stoma was found in the other two cases, and one proximal stoma was stenotic. Seven loop colostomies with faecal impaction of the distal bowel were observed. Twenty-one stomas were 'double-barrel' or too close to each other to apply a bag. These devices are not readily available in Sudan. Many families cannot afford them and cover stomas with a rudimentary bandage; the passage of stools into the distal stoma is not prevented. In conclusion, 38% of the colostomies observed did not satisfy the currently recommended criteria.

### Imaging

A contrast study of the distal loop was available in 47 out of 66 cases only. Poor quality of these studies was a common problem. Loopograms at the GNCPS are usually not supervised by a paediatric surgeon but are committed to an X-ray unit external to the centre operated by technicians lacking specific expertise. The loopogram confirmed a short distal loop secondary to intraoperative sigmoid twisting, in ten cases, and to a low sigmoid stoma in five cases. Low-pressure injection of contrast and presence of stools or faecal debris were responsible for missing recto-urinary fistula in males.

**Table 1: Ninety-four patients with anorectal malformation – distribution for type of anomaly (Krickbeck classification)**

Type of ARM	Number of cases
Perineal fistula	5 (4 males; 1 female)
Rectobulbar fistula	34
Rectoprostatic fistula	2
Rectovesical fistula	4
Rectovestibular fistula	36
Cloaca	7
No fistula	2 (1 female; 1 male)
Rectal atresia	2 male
H-type fistula	2 female

ARM: Anorectal malformation

**Table 2: Management and outcome of 94 patients with anorectal malformations referred to the Gezira National Centre of Pediatric Surgery**

Procedures planned	Group A: 80 ARMs not operated		Group B: 14 post-PSARP complications (2 with colostomy)	
	66 with colostomy (38% unsuitable)		14 without colostomy (1 anteriorised anus no surgery)	
	17 (25%) colostomy revisions 9 short loop 5 loop colostomy 2 prolapse 1 stenosis	PSARP 49	Colostomy 11	2 perineal fistula (no colostomy)
	PSARP 17		13 PSARP	
				1 severe anal stenosis 1 prolapsed mucosa 12 poor incontinence 2 surgical revisions (prolapse and stenosis) 3 Malone appendicostomies 9 PSARP redo (7 after a new colostomy)
Procedures done	Group A		Group B	Total
Colostomy	12/17 (revision)	-	9/11	24/35
PSARP	6/17 (1 TUGM)	40/49 (1 TUGM)	7/13	57/88
Complications	7 PSARP wound infections		-	2 PSARP wound infections 9/57 (16%)
Follow-up and results				
Stoma closure	3/6	18/40	3/7	26/57 (45%)
Outcome	1 poor continence	4 mucosal prolapse 2 anal stenosis	1 mucosal prolapse	-

PSARP: Posterior sagittal anorectoplasty, TUGM: Total urogenital mobilisation, ARM: Anorectal malformation

### Operational plan

Table 2 shows an individual plan which was made based on age, type of ARMs, associated diseases, type of stoma and previous surgery. Colostomy revision was scheduled for 17 out of 66 patients in Group A (9 for a short distal loop, 5 for a loop colostomy, 1 for stenosis and 2 for prolapse). A divided colostomy was planned for undiverted patients except for two recto perineal fistulas eligible for a one-stage procedure. Female patients were never diverted before 1 year of age if evacuation through the rectal fistula was warranted. Except for one girl with an anteriorly displaced anus, not requiring treatment, PSARP was scheduled for all cases in Group A associated with total urogenital mobilisation (TUGM) in six 'short common-channel' Cloaca cases. Permanent faecal and urinary diversion and vaginal reconstruction were planned in a long common-channel Cloaca case. PSARP re-do was advised in 9/14 cases in Group B for an anal misplacement associated with poor continence. Two already had a colostomy, and seven needed a new stoma. In three patients where PSARP re-doing was not feasible due to severe scarring of perineum and muscular complex, a Malone appendicostomy was planned. One prolapse and one stenosis responded to a limited surgical treatment without PSARP re-do. In conclusion, we scheduled 35 patients for primary or re-do colostomy and 88 patients for a primary or re-do PSARP.

### Pre-operative nursing

Among cases which had already a colostomy, 'double-barrel' or too close stomas were frequently observed. There was an associated risk of distal loop faecal contamination, and accurate cleaning by antegrade washout was sometimes challenging to achieve in order to prevent post-operative surgical-site infections.

### Operational activity

Twenty-five colostomies (12 re-do, 13 primaries) and 57 PSARP (4 re-do, 53 primaries) were performed (TAB II). PSARP was associated with a TUGM in two Cloaca cases. An abdominal perineal approach was required in two cases of rectovesical fistula. Thirty-one patients were on the waiting list for PSARP (5 re-dos). Twelve had colostomy already done, but three among them were still missing readmission after 1½ years from the diversion. To minimise the risks of losses from follow-up, we started doing colostomies 2 weeks only before PSARP also to reduce the burden of multiple admissions on many low-income families and many long-term, stoma-related problems.

Paediatric surgical registrars did about 30% of the procedures following rigorous technical rules and using a muscle stimulator, donated to the GNCPS by the AICS and not available before. General surgical registrars, rotating at the GNCPS, were trained to correctly confectioning a divided colostomy before referring cases to a paediatric surgical specialist.

## Complications

Surgical-site infections occurred after 9/57 PSARP (15.7%); two of them after a re-do. Three wound dehiscence required surgical revision. A secondary procedure was needed in three boys after the urethral catheter was accidentally removed, followed by urinary leakage. An abdominal incisional hernia needed repair, and one death was registered after colostomy closure due to anastomotic dehiscence and sepsis. No significant correlations were found between perineal infections and age, gender and type of ARM. Although infections were more frequent among PSARP cases with a colostomy on referral (8/47; 17%) respect to those with a colostomy done after referral with a correct technique (1/10), the correlation was not significant.

## Follow-up and outcome

Post-PSARP dilatations by Hegar started 2 weeks after surgery. There were no outpatient clinics for ARMs at the GNCPS and many patients were at risk to be lost from follow-up. Not enough time was spent warning mothers on preventing post-operative stenosis by anal dilatations. Many families lived too far from hospitals, and used homemade dilators (catheters, finger) with uncertain results<sup>[6]</sup> as personal sets of dilators were not affordable. Twenty sets of plastic Hegar sounds, supplied to the GNCPS by the AICS, could be rented by families by payment of a minimum refundable deposit. A nurse was appointed to distribute sets and collect them after use, to follow mothers by telephone, to keep records updated, to check compliance to the dilatation protocol and to schedule colostomy closure in due time. During the period of observation, the quality and length of post-operative follow-up resulted in being satisfactory in about half of the 57 PSARP cases (average time 6 months; range, 3 months–2 years). Stoma closure after PSARP was possible only in 26/57 (47%) cases after an average time of 3 months and 9 days (range 1–15 months). These figures reflect difficulties in contacting and recalling families living in rural areas, with problems in travelling and accessing hospital care. Two anal stenosis and four prolapses required surgical management before stoma closure. One case of poor continence was recorded in a child with Down syndrome, and a bowel management programme has been proposed to the family.

## DISCUSSION

Since the 1980s, the PSARP approach<sup>[5]</sup> has changed the philosophy of management for ARMs. A growing interest for it is developing over the sub-Saharan Africa, although the small number of paediatric surgical specialists fights to meet the needs of the paediatric population.<sup>[3-7]</sup>

The relevance of ARMs in Africa has been already emphasised as figures rate them as the most frequent (57%–67%) neonatal intestinal obstruction and the most common (up to 64%) reason for neonatal surgical referral.<sup>[3-10]</sup>

The role played by ethnicity and genetic influences on this higher incidence of ARMs with respect to other defects,

compared with other continents, is still debated.<sup>[11]</sup> Hidden mortality could be speculated among neonates with more severe malformations, leading to a relatively higher incidence of ARMs in the sub-Saharan Africa.<sup>[12]</sup> Delayed referral from rural areas can reduce the number of male cases who can die before an emergency colostomy is done.<sup>[13]</sup> Girls with a vestibular fistula may remain undiscovered until adolescence without any sign of obstruction.<sup>[4]</sup> In addition, cases with life-threatening abnormalities may be lost before referral, keeping their incidence below the expected rate.<sup>[1-14]</sup> Female prevalence and low rate of associated anomalies registered among our cases are consistent with these observations. Ultrasound screening for foetal abnormalities is unknown in Sudan. Cardiovascular or urinary abnormalities frequently associated with ARMs are often missed and occasionally discovered on hospital admission.

Despite the high relevance of ARMs in the sub-Saharan Africa, an updated approach could be introduced only in recent times, but it remains confined to major centres in urban contexts, where few paediatric surgeons and well-equipped facilities offer an adequate standard of care to a limited number of patients. A strong effort is requested in terms of economic resources, infrastructures and trained workforce to extend the benefits of modern surgery to a more significant number of children.<sup>[15]</sup> PSARP approach may so far be constrained even in some tertiary centres. Low-resource context hampers strict adherence to the recommended criteria for a correct staged approach. A high rate of complications or failures may, therefore, be expected.<sup>[16-19]</sup>

We divided our cases observed at the GNCPS into two groups. The first composed of untreated ARM cases with or without a diverting colostomy (Group A). The second comprised cases referred to after a previous PSARP (Group B) where we limited re-doing to those cases with a misplaced anus, which could reasonably benefit from surgical repositioning under electric muscular stimulation. In both groups, the first concern was on colostomy, which is a major medical and social problem in the sub-Saharan Africa.<sup>[20]</sup> There is a low level of acceptance for stoma among people living in rural areas, especially when access to definitive surgery is delayed due to poverty or lack of tertiary facilities. Before a PSARP can be performed, a divided stoma, following a rigorous set of rules, must be recommended.<sup>[21]</sup> The short distance between stomas, as frequently observed at the GNCPS, is associated with stools entering the distal loop, leading to faecal impaction. Although 38% of the colostomies did not strictly respect the recommended standard, we limited re-confectioning before PSARP to 17/66 cases only (25%) if a short distal loop interfered with rectal mobilisation or when a prolapse or stenosis was present or when loop stomas were found. Pre-operative loopograms were of scarce help, in most of the cases, due to the scarce familiarity of residents, with imaging associated with inadequate X-ray facilities.

Lack of supervision on residents by the very few consultants acting at the centre could explain the poor quality of colostomies observed at the GNCPS. Some of them were also done in a non-specialist referring hospital. The stoma complications seen at GNCPS (prolapse, inversion, etc.) have been rated from 81%<sup>[22]</sup> to 26%<sup>[23]</sup> in different African series. Significant mortality, up to 25%, may be associated with colostomy in small children.<sup>[14]</sup> Stoma-related problems can explain the favour for a 'one-step' PSARP approach.<sup>[24-27]</sup> According to our experience, the risk of wound-site infection accompanying PSARP without colostomy is high. If intravenous nutritional support is not available and nursing care remains defective, a divided stoma is still the best practice even in a well-resourced context. We have no reason to let our guard down where the local context is less favourable.<sup>[16-21]</sup> Most of these stoma-related problems can be prevented, in our opinion, by better surgical training instead of recurring to 'one-step' solutions.

Fifty-seven patients could benefit from PSARP until now. The perineal surgical-site infection rate was lower among those cases who had their first colostomy confectioned according to proper technical rules, but bedside wound contamination remains a problem at GNCPS due to a lack of bedding sheets and poor nursing. Infection is a commonly reported complication among African series with variable incidence rates of 41%<sup>[18]</sup> and 21%<sup>[28]</sup> and possible solutions to minimise the risk of complications have been proposed.<sup>[29]</sup> This risk is higher among HIV-exposed cases that need strict perioperative protocols.<sup>[30]</sup>

Post-PSARP anal dilatations and bowel management are a critical issue for GNCPS as in many paediatric surgical centres.<sup>[4-16]</sup> Patients may be lost from follow-up and never come for the definitive treatment after colostomy due to economic reasons and inadequate transport facilities. It happened in a few of our cases. The compliance to anal dilatations following PSARP and even after bowel recanalisation is not always warranted, and the risk of stenosis is high. Supplying a set of plastic Hegar dilators to each family is a possible solution, but without community social workers, the review of patients in their place of residence is somewhat tricky. Since the beginning of the present programme, only 26 patients (46%) could have their colostomy closed to consent their final outcome being thoroughly evaluated. Poor continence was registered in a child with Down syndrome. PSARP approach to ARMs requires rigorous staging, meticulous technique, electric muscle stimulation, appropriate pre- and post-operative care and long-term follow-up. Without these conditions, poor outcome after PSARP is behind the corner. If incontinence results from an unconventional approach, poor technique and inadequate post-operative care, the statement that '...a permanent colostomy is preferable to a "perineal stoma"...', must be kept in mind.<sup>[16]</sup>

## CONCLUSION

Lesson learned from the ongoing programme confirms that approach to ARMs may be strongly constrained in some sub-Saharan countries, even at a tertiary centre, but not exclusively by lack of infrastructures and poverty. A continuous brain drain of trained health providers reduces the number of tutors available for postgraduate education. In a continent like Africa where the number of surgeons is low, training of a large number of residents is the only way to fill the gap. Unfortunately, the expansion in the number of trainees in an understaffed teaching context risks to keep the quality of training below acceptable standards<sup>[31]</sup> and favour the acquisition of technical skills over the aptitudes for auditing and clinical research. We committed our academic partnership not only to strengthen the educational resources and the teaching activities of the GNCPS for the new specialists but also to provide general surgical residents a basic paediatric knowledge. The number of pediatric surgical residents is increasing in Sudan to cope with the largely unmet needs of the country, but brain drain curtails the number of specialists towards better job opportunities abroad. As far as resources for children's surgery in Africa remain low, an alternative can be offered by trained general surgeons delivering primary surgical care for neonates and children in rural and district hospitals and providing a safe referral to specialist centres.<sup>[32]</sup> As far as trained nursing staff is concerned, a scarce interaction with physicians and low wages results in scarce motivation and a lack of self-recognition. In this context, academic international partnerships may play a key role, but without more attention to the recruitment, motivation and retention of trained healthcare professionals, few can be done to improve performance and upgrade the standard of care.

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## Conflicts of interest

There are no conflicts of interest.

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