Unique challenges in managing pediatric colorectal diseases in under-resourced areas: context-aware adaptive responses from short-term surgical outreach visits

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Abstract

Pediatric Colorectal Diseases (PCRD), mainly Anorectal Malformations (ARM) and Hirschsprung's Disease, are a major issue in Sub-Saharan Africa (SSA). Even with advances in healthcare facilities and global health initiatives, most of children living in low resources SSA lack of specialist pediatric surgical facilities. Improved healthcare access is needed to manage PCRD in these locations, to prevent and reduce missing or delayed diagnosis, early mismanagement by inexperienced health practitioners, and barriers to corrective therapy and long-term follow-up. A retrospective analysis of data from three SSA hospitals shows that international surgical outreach visits helped increase capacity. Along one hundred twenty-four weeks of staggered surgical outreach visits 174 ARM and 64 highly suspected HSCR cases were collected. The study evaluates 152 ARM and 59 HSCR patients who had not been treated before. Those who came after an unsuccessful treatment elsewhere were not included. Management, clinical course, complications and results are reported. Focus is on context-aware adaptive surgery and sustainable solutions to improve outcomes and quality of life for those children, discussing long-term follow-up options and results. The local context has a substantial impact on epidemiology, demographics, and presentation compared to high-resource countries. Intestinal stomas done at a primary health facility level presented at referral to our Hospital with 25% complication rate. Due to social, economical, and transit issues, only 108 ARM and 41 HSCR could finally receive a corrective treatment by the outreach visiting teams. Complications (9.1%) were controlled using adaptive solutions. Only one surgical fatality occurred. A limited proportion of patients (46% ARM, 31% HSCR) attended a regular follow up schedule for one year or more, and finding those lost in wide rural areas was difficult. Successful and comprehensive PCRD management in under-resourced SSA requires better training at the primary health level on early recognition and correct, first surgical approach, together with a referral network to specialist facilities for further treatment. Surgical short-term outreach trips can boost local capability in under-resourced areas. The research of adaptive and sustainable surgical solutions to reduce hospital stay and staged treatments time for PCRD must be emphasised. Nevertheless financial and logistical constraints still challenge post-discharge monitoring and follow-up, which remain crucial for long-term outcome.

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Introduction

Pediatric Colorectal Diseases (PCRD) represent for Sub-Saharan Africa (SSA) a significant portion of the surgical burden of congenital abnormalities and the most frequent cause[1] of intestinal obstruction in children. Despite the establishment of new tertiary specialist facilities in central urban areas in recent years and the contribution of global health initiatives, advanced pediatric surgery remains inaccessible to many children, approximately half of the resident population, in vast low-resource regions of SSA.

Therefore, Anorectal Malformations (ARM) and Hirschsprung's Disease (HSCR), both of which require an early, qualified approach and advanced surgical skills, risk missed or delayed treatment and fall under the care of untrained health providers, with potential mismanagement and poor outcomes.[3]

Although their limited "stay on the target", international surgical outreach visits for children can provide valuable assistance and promote capacity building. Nevertheless, visiting healthcare providers requires understanding local constraints and managing PCRD within a framework of context-aware, adaptive surgery to achieve long-term, satisfactory clinical results and a good quality of life. This document examines these issues based on the authors' experience in three SSA health Institutions.

Materials and Methods

Aim

This paper aims to investigate the management of PCRD in under-resourced areas of SSA focusing on: i) the impact of the context on epidemiology and demographics; ii) how an early approach at a primary health level influences cases' presentation for surgical correction; iii) local constraints accessing corrective treatment; iv) barriers to post-discharge monitoring and follow-up; v) possible context-aware adaptive responses.

Study setting

A retrospective analysis of prospectively collected data from the records of all patients, from birth to 18 years, with a PCRD (ARM and HSCR) observed and treated between 2011 and 2024 in three SSA hospitals during one hundred twenty-four weeks of staggered surgical outreach visits by thirty-nine teams from Pediatric Surgical Italian Institutions. The hospitals were: i) Orotta National Referral Hospital (ONRH) and the 300-bed annexe of Mekane Hiwot Pediatric Hospital in Asmara; it serves as Eritrea's central public health institution, mainly in an urban catchment area, but it receives complex cases from nearby rural areas (42 weeks); ii) the Gezira National Centre of Pediatric Surgery (GNCPS) in Wad Medani is a university-affiliated institution in the Sudanese State of Gezira, serving patients from both urban and rural contexts (30 weeks); iii) Consolata Hospital Ikonda (CHI) is a charitable institution that serves as a regional referral hospital in the Makete district of the Tanzania Highlands, far from the main metropolitan areas (52 weeks)

The primary purpose of outreach visits was educational. Team members were intentionally limited in number to enforce a complete integration model with local staff. The primary objective was to enhance capacity through on-the-job training and coaching, to share knowledge and experience, and to implement a sustainable standard of care by optimising the use of available resources.

Data collection

Data were retrieved from hospital records regarding demographics, presentation, diagnostic workup, management, complications, follow-up, and outcomes of ARM and HSCR cases.

Data entry

The GraphPad Prism 8.4.0 statistical package (San Diego, CA, USA) was used for data entry and analysis of frequency distributions and percentages.

Ethical standards

The study was conducted by the 1964 Helsinki Declaration and its subsequent amendments, or comparable ethical standards, as established by local surgeons and the outreach team.

Results

Case series

One hundred and seventy-four ARM cases (6.8%) were recorded during the study period among 2543 pediatric surgical admissions (including urological cases). Only 152 patients with an uncorrected ARM were considered in the study. An additional twenty-two cases (M/F 14/8) aged between 5 months and ten years (mean 4.8 years) referred after a failed previous corrective surgery in other Hospitals were excluded.

Sixty-four patients with a suspected diagnosis of HSCR were admitted in the same period. They were 2.7% of all surgical admissions. Their symptoms varied from an acute abdomen to a severe refractory chronic constipation. Clinical history from birth was not always detailed or available. Five children came after having been treated with an unsuccessful pull-through procedure at another hospital and were not included in the study.

Demographics

The male-to-female ratio among 152 ARM cases was 84/90, indicating a slightly higher prevalence in females. The mean age was 2.1 years (Median 1 year, range 3 days to 18 years). Among the fifty-nine patients suspected of having HSCR, the male-to-female ratio was 46:13, suggesting a significantly higher prevalence in males. The mean age at admission was 3 years (median, 2 years; range, 1 month to 13 years).

Presentation

A colostomy was present in 103 children with ARM (67%). This group included fifty-nine males aged between one month and 16.9 years (mean of 1.7 years, median 1 year) and forty-three females between three months and 18 years (mean of 1.7 years, median 1 year). Diversions had been generally made for obstruction as an emergency at a primary level of care by non-pediatric surgeons. Thirty stomas (29.1%) presented with functional problems or associated complications. The sigmoid was the preferred site for diversion, and a loop colostomy was the most frequently adopted technique, followed by divided stomas. Poor diversion from stools was the main complication associated with loop colostomy; severe fecal impaction in the distal loop was found in four cases. Prolapse was not significantly associated with specific stomas but appeared more closely related to the bowel segment selected, such as the transverse colon. In about one out of four sigmoid colostomies (26%), the position of the stoma had been inverted by the surgeon, and the distal loop was at risk of being less mobile to reach the pelvis without tension at a Posterior Sagittal Anorectoplasty (PSARP). Nine per cent of sigmoid loop colostomies were too low, resulting in a distal loop that was too short.

Forty children (67.7%) admitted for a suspected HSCR had a levelling colostomy done in an emergency before referral for an acute obstruction; in one of our hospitals, all the patients referred already had a stoma. No investigation had been conducted to confirm HSCR and the length of the aganglionic tract; the surgeon had chosen the diversion level based solely on the colonic caliber. Apart from two on the right colon, all the stomas were located on the sigmoid; 25 cases (62.5%) had a loop colostomy, complicated by prolapse in three and stenosis in two. Four divided and ten terminal

colostomies (one stenotic) were also recorded. The rate of stoma-related complications among HSCR patients was lower (15%) than among ARM cases.

As stoma bags are frequently not affordable or available in these areas, parents often resort to homemade solutions, using clothes, commercial diapers, or cloth napkins to cover the stoma and collect stools. However, these solutions did not adequately protect the skin, resulting in peristomal erosions, fungal infections, and bleeding of the exposed mucosa in our patients. Prolapse was often associated with mucosal erosions and bleeding, further complicating the condition. Table 1 reports the types of stomas observed and their related problems.

Forty-nine children with ARM (M/F 11/38) came without intestinal diversion. Three male neonates only were admitted for an obstruction with an acute abdominal distension. Another eight males, aged from one month to one year, had been passing stool since birth through a Rectoperineal fistula. Females without a stoma presented with a rectovestibular fistula (thirty-six) or a Cloaca (two) and came to the mean age of 1.7 years (median 7 months, range 1 month to 18 years).

Nineteen cases were admitted for a suspect HSCR without a colostomy. Three males and one female only presented in an emergency in the first four months of life, with a clear history of delayed emission of meconium at birth and an acute abdomen (one with a colonic perforation). Another fifteen patients complained of severe refractory chronic constipation and chronic abdominal distension, which was currently relieved by the mother with repeated enemas. Malnutrition was most frequently associated. Clinical history from birth was not always detailed or available from their families.

Diagnosis

The diagnostic workups were significantly influenced by the resources available in each institution. Only two hospitals had Ultrasound (US) and complete X-ray equipment. In one case, the preoperative assessment had to be limited, in most cases, to a clinical examination. All three hospitals in this study had access to a pathology laboratory. However, frozen intraoperative diagnosis sections were unavailable, highlighting the need for improved resources and technology in the diagnosis process.

Whenever possible, associated malformations to ARM (cardiovascular, urological, gastrointestinal) and chromosomal defects were investigated by systematic screening, including renal, spinal, and cardiac imaging, and were found in only nine cases (5.9%). A cross-table lateral radiograph was always available in two Hospitals for a high-pressure distal colostogram to define the type of rectourethral fistula in males. In a third Institution, a colostogram could be performed only sporadically, and recto-urinary fistulas in males were identified intraoperatively.

In forty cases referred for a suspected HSCR and with a colostomy, a full-thickness biopsy for ganglia was performed at the level of the rectum and stoma. Rectal aganglionosis was confirmed in thirty-three out of forty (82.5%). A distal loop gram was also performed in all patients diverted before admission, excluding cases with a terminal colostomy, to confirm the stoma's level in the colon and observe the loop's aspect and length.

The diagnosis of aganglionosis was confirmed in a perforated neonate, both by rectal and intraoperative colonic biopsies, during the emergency diversion on admission. In another three infants with an acute obstruction and needing urgent levelling colostomy, a preoperative plain abdominal X-ray was highly suggestive of HSCR, and intraoperative and rectal biopsies confirmed the diagnosis. Another four undiverted patients were investigated by a plain abdominal X-ray and a barium enema, which identified a transition segment at the mid-sigmoid level. A full-thickness rectal biopsy confirmed the aganglionosis, which was also evident in multiple colonic biopsies taken during the levelling loop colostomy construction. In another eleven non-diverted cases with chronic refractory constipation (M/F 9/2, mean age 4 years, median 3.5 years, range 4 months-9 years), a barium enema suggested short segment aganglionosis or achalasia with a dilated stool-loaded rectal ampulla. We opted in these cases for a cleaning enema followed by a posterior sphincterotomy and rectal myotomy. The purpose was therapeutic, in addition to the diagnostic procedure of rectal biopsy. Aganglia was confirmed in seven out of eleven of them (63%).

Access to corrective surgery

Table 2 outlines our cases with ARM or HSCR from the initial admission. Fitness for corrective surgery (PSARP) was assessed for all ARM patients (state of nutrition, associated abnormalities or acquired diseases) together with the capabilities of families to comply with the main steps of treatment (scheduled admissions,

Table 1. Type, site, complications.

ARM	34 DIVIDED 17 Complicated (50%)		8 DOUBLE BARRELL	61 LOOP 13 Complicated (21%)	
95 SIGMOID Complications 27%	34	2 prolapse 1 proximal stoma stenosed 9 inverted stomas 5 short distal loops	8	53	5 short distal loops 4 prolapse
6 TRANSVERSE COLON Complications 33%		-	-	6	2 prolapse
2 RIGHT COLONIC FLEXURE Complications 100%		-	-	2	2 prolapse
HSCR	**4 DIVIDED**		**11 TERMINAL 1 Complicated (9%)**	**25 LOOP 5 Complicated (20%)**	
38 SIGMOID Complications 15%		3	10 1 stenosis	25	3 prolapse 2 stenosis
1 RIGHT COLONIC FLEXURE		1			
1 CECUM			1		

colostomy management, postoperative dilatations). Four children (M/F 2/2) with congenital cardiopathies associated with two cases of Down syndrome were excluded from further corrective surgery. All could pass stools through a stoma or a large rectovestibular fistula. For another 29 patients (M/F 10/19), the families failed to attend the scheduled admission for surgery or postponed treatment indefinitely. Eighteen among them had been referred already with a stoma; the other children, without a stoma, could pass stools through a Recto-vestibular fistula, a Cloaca, a Recto-perineal fistula, and an H-type fistula.

Twenty cases out of 82 (24%) with a stoma that did not respond to the criteria for a safe PSARP procedure[4] needed a redo or revision procedure. Twisting of the sigmoid with a consequent inverted stoma position, sometimes associated with a short distal loop, was the indication in 40% of re-interventions, followed by prolapse (30%) or stenosis of the proximal stoma (30%). To save many children a surgical revision, the distal stoma of loop and double-barrel colostomies, which was a possible risk of post-PSARP perineal contamination, was currently closed before the corrective procedure by a purse string suture after an accurate antegrade washout.

Three obstructed male neonates with ARM needed an emergency divided colostomy on admission. A divided sigmoid stoma was also constructed in 24 females at a mean age of 1.3 years (range: 1 month to 7 years) for rectovestibular fistulas (22 cases), H-type fistulas (2 cases), and one short-channel Cloaca. No postoperative problems arose after the colostomy was of the divided type. All patients were supplied with stoma bags by the hospital during their stay and sometimes at discharge to prevent wound infections and peristomal skin damage.

A PSARP could be performed in 108 cases among 119 (90%) who accepted the corrective treatment. Eleven cases are still on the waiting list. Some of them omitted coming due to economic or logistic reasons, and many were impossible to contact.

Seven perineal fistulas (M/F 4/3) had a one-step PSARP without diversion within the first three months of life. In another three malnourished, underweight males with a perineal fistula, a preliminary diversion was preferred by the surgeons to a one-step approach because these patients demonstrate a higher risk of impaired wound healing, skin breakdown, and wound dehiscence.[5]

A PSARP under stoma protection was performed in 101 patients, including three total urogenital mobilisations for Cloaca (one long and two short common channels). Starting in 2018, two of our hospitals received a muscle electric stimulator. The electrocautery set with a low voltage was used as an alternative to identify the muscular complex.

A technical modification of the original PSARP was introduced, when technically possible, in some patients to protect perineal wounds from anal mucous discharge.[6] It was a significant problem in an area affected by a shortage of nursing care and cleaning devices. The modification consisted of leaving a long rectal stump anchored by eight Vicryl stitches to the perineal skin at the anal opening and closed by a double purse-string suture. This procedure helped to keep the perineal area clean and to prevent local infections and suture disruption. The patients were discharged from the Hospital 8-10 days after surgery, and resection of the rectal stump at perineal skin level was performed two weeks after, using a simple thermocautery under mild sedation.

A colostomy reversal was planned for seven diverted patients admitted for suspected HSCR after the aganglionosis was not confirmed by rectal biopsy. For another eleven cases with severe chronic refractory constipation and imaging of short-segment obstruction, we opted for a cleaning enema followed by a posterior sphincterotomy and rectal myotomy for diagnostic and therapeutic purposes.

As frozen sections were unavailable in all Hospitals to consent to a one-step pull-through procedure, an elective sigmoid loop colostomy was performed on eight children after a barium enema demonstrative of a rectosigmoid transition zone and a positive rectal biopsy. During the diverting procedure, the surgeon took multiple seromuscular biopsies from the distal colon to be examined later and to delimitate the length of the distal aganglionic tract. Additionally, the revision of complicated colostomies, which were performed prior to referral, provided the opportunity to take multiple biopsies of the distal colon.

To avoid a third surgical procedure for stoma reversal, which carries possible risks and higher family costs, we adopted a direct

Table 2. Management flowchart of ARM and HSCR cases.

174 ARM	84 Males	90 Females	64 Suspected HSCR	49 Males	15 Females
Previous correction failures (not included in the study)	14	8	Previous correction failures (not included in the study)	3	2
152 Included in the study	70 Mean age 1.5 yrs Median 10 mo	82 Mean age 2yrs Median 11 mo	59 Included in the study	46 Mean age 3 yrs Median 2 yrs	13 Mean age 3 yrs Median 2 yrs
Lost/Unfit for surgery	13	20	7 Lost HSCR non-confirmed by rectal biopsy	6 9	1 2
Scheduled for PSARP	57	62	Scheduled for surgery	31	10
108 PSARP (7 one-step)	50 Mean age 1.7 yrs Median 1 yr	58 Mean age 2.1 yrs Median 1 yr	34 Two Steps PULL-TROUGH De La Torre 15 Duhamel 15 Soave 4	26 Mean age 2.9 yrs Median 2 yrs	8 Mean age 5 yrs Median 2 yrs
STOMA REVERSE 71/101 cases – 70% Interval to stoma reverse	Mean interval 6 mo Median 3.8 mo		7 SPHINCTEROTOMY Short Segment Aganglia	5	2
Follow-up ≥1 yr 49/108 (46%)				Follow-up ≥1 yr 13/41 (31%)	

pull-through solution for the functioning stoma, where the presence of ganglia had already been warranted.[7]

A Duhamel pull-through (15 cases) was preferred for seven children with a terminal colostomy and a residual short rectal stump. The side-to-side anastomosis was accomplished using a linear stapler (Auto Suture PolyGia 75 Single-Use Stapler with Absorbable Staples). A classic Soave procedure was performed in four cases, and a De La Torre transanal endorectal pull-through in fifteen.

We began with a single-layer provisional colostomy closure to temporarily restore bowel continuity. All mesocolic adhesions to the left parietal peritoneum up to the splenic flexure or above were dissected to consent to a tension-free pull-through, especially in cases with a terminal colostomy. When a long distal loop was present, it was prepared for resection, and its vascular supply was ligated down to the level where separation of the seromuscular from the mucosal layer of the rectum should start for the Soave procedure. In the case of the De la Torre procedure, vessels were ligated until the peritoneal reflection was reached. The de-vascularised a-ganglionic segment was then resected two centimeters above the previous stoma level, as the sutured stoma site appeared from the anus by transanal traction. A standard Soave Boley coloanal anastomosis was performed to conclude the procedure.

Postoperative course and follow-up

There were no anesthesiologic complications. Eighteen cases (16%) had an early wound infection of various degrees; ten (9.2%) also presented a wound dehiscence, which required a surgical revision under anaesthesia. The accidental removal of the bladder catheter in two cases caused a temporary urinary leakage from the perineal wound, which required a new catheter in place for one month, associated, in one case, with a temporary cystostomy.

Colostomy reversal after PSARP was usually performed at a mean distance of approximately six months from the corrective procedure, provided that the anal dilatation program achieved the expected results. Some reversals required a longer wait, up to one year. A significant number of patients (37 out of 108, 34%) missed until now the stoma reversal in the expected time due to the inability to cope with the follow-up schedule or to respect the anal dilatations protocol. Financial and transportation difficulties in long-distance travel were also influential in the delay of stoma reversal.

Colostomy closure was often problematic. Poor prevention of intraoperative contamination, infectious comorbidities such as HIV infection and malnutrition, and insufficient nursing care contributed to infections of various degrees in 20 cases out of 71 (28%), with associated skin suture disruption and prolonged hospital stay.

Besides irregular attendance at the postoperative schedule, devices to follow the dilatation protocol at home, which are often necessary starting 2-3 weeks after the procedure, are often unavailable. Homemade solutions, such as rectal tubes of the proper size and lubricated parents' fingers, may not always be ideal for the purpose. Hegar dilator sets are expensive and unavailable in peripheral regions. A solution introduced in our hospitals was to supply families before discharge by paying a small deposit, along with a disposable cervical bougie set ranging from caliber 7 to 13. Families were taught to increase the caliber of the sound progressively. For older patients, lubricated rectal tubes of appropriate size were dispensed.

A regular follow-up of more than one year after a post-PSARP colostomy reversal could only be documented in 46% of cases. Constipation due to a tight or fibrotic anal ring was observed in about two-thirds of these patients. Poor fecal control or significant soiling was observed in five cases, one with an associated Down Syndrome. Postoperative anal stenosis required a strictureplasty in ten cases.

One two-year-old female patient with HSCR died after a Duhamel procedure for an Atrio-Ventricular Nodal Reentrant Tachycardia, not responding to the locally available medications. We had two limited anastomotic leaks and cuff abscesses after a De La Torre and one para-anastomotic abscess after a Duhamel procedure requiring an emergency temporary colostomy. One of the patients died of sepsis, and two recovered, and the stoma could be closed a few months later without sequelae. Another patient needed a surgical revision for obstruction due to twisting of the pulled-through bowel during a De La Torre procedure. One para-anastomotic abscess after Duhamel could be treated conservatively. Three patients developed anastomotic stenosis after two De La Torre and one Duhamel procedure, which responded to dilatations. One late death was recorded for an acute episode of enterocolitis after a Soave procedure in a six-month-old boy. All other patients had regular bowel movements six months after surgery, but regular follow-up records at 1 year from pull-through were available in only 31%. Sphincterotomy relieved constipation in all patients who had a short segment obstruction, either with a ganglionic or with a ganglionic rectal biopsy. Only one of these patients required a second procedure after an incomplete sphincterotomy.

Discussion

The impact of the context on the epidemiology and demographics of PCRD

The cumulative load of ARM (3.11 for 10,000 live births) and HSCR (1.34 for 10,000 live births) makes PCRD the most prevalent among major congenital anomalies worldwide.[8] Although the incidence of PCRD does not seem different in High-Income Countries (HIC),[11-14] their recorded rate, epidemiology and demographics in SSA[15,16] may result differently as influenced by more births and children than in HIC[9] and many without access to pediatric surgical services,[10] missed or delayed diagnoses related to the high number of deliveries outside health facilities,[17] scarce level of awareness and recognition among primary health providers in rural areas,[18] hidden mortality in the first days of life from associated lethal anomalies[19] unable to access health facilities due to economic[20] or transport problems.[21,22]

Females with ARM with a rectovestibular fistula may remain undiagnosed for a long time beyond puberty.[23] Male neonates with a rectourinary fistula who require early diversion to survive, Gender-related prevalence, age at referral, and survival of ARM in SSA results influenced[24] as reflected in our series. The male-to-female ratio in our ARM group, which is estimated at 1.7 in HIC,[25] was inverted to 0.8.

The mean age at referral was not significantly different between males and females, but it was much higher than is usually recorded in HIC, where most ARM patients are newborns.[26] There were only three neonatal referrals in our series, all of whom were males.

Male patients were fewer in number than females but had a higher proportion of colostomies on referral (84% vs 52%, $p < 0.05$). Male survival mostly depends on an emergency colostomy.[27,28] Many losses can be attributed to a delay in diagnosis or seeking care during the neonatal period. A delay in referral with the consequent loss of the more fragile patients could explain the congenital malformation rate associated with ARM far below (5.9%) the expected (up to 75%).[29]

If we compare the distribution of different types of ARM[30] among our operated cases (Table 3) with that from The European Anorectal Malformation Network (ARM-Net) patient registry,[31] the incidence of perineal fistulas among our and other African series (8%-15%) is lower ($p<.05$) than among European patients

(41%).[13,27] It could be due to a delayed or omitted referral of anomalies, which have a minor impact on everyday life.[32] Rare regional variants, such as H-type fistulas, Rectouretroperineal fistulas, and rectal atresia, are represented among SSA cases and in our series.[33]

HSCR management shares the same constraints as ARM [9]. Neonatal cases with early obstruction are a minority in SSA compared to HIC (20-40% vs 90%, 6.7% in our study).[34,35] There is a prevalence, among referrals for HSCR, of patients with a history of severe chronic constipation, which is a highly sensitive symptom, but it is not specific.[13] The mean age at the referral of our non-neonatal cases was three years (median 2 yrs, range 3 months-13 yrs), and 20% finally resulted in being chronically constipated without aganglionosis. A diversion had been required at a primary health level for an acutely distended abdomen in seven cases. In conclusion, the low awareness associated with hidden neonatal deaths, the high prevalence in African children of chronic constipation extensively treated with traditional remedies, and the frequent recourse to diversion at a primary health level without any diagnostic evidence contribute all to influence the mode of presentation and the estimated prevalence rate of HSCR in SSA.[36-39]

The impact of the primary health-level approach

The prevalence of missed or delayed referrals and mismanagement of PCRD in SSA is commonly attributed to the inadequacy of primary care.[40] Our figures confirm this issue, emphasising the crucial role of healthcare professionals in the primary care system and the need for its improvement.

Twenty-two patients with ARM and 5 with HSCR had a previous unsuccessful corrective surgery in another hospital. Most of them had been operated in a non-specialist context by non-pediatric surgeons. They usually perform successful pediatric lifesaving procedures in rural SSA. However, whenever they attempt to correct complex anomalies, they expose patients with PCRD to severe complications or compromise the results of further corrective surgeries.[41,42] The same can happen when specialist procedures are done by visiting general surgeons without pediatric experience on the basis that in an under-resourced context, "doing something is better than doing nothing" and "children are small adults".

One hundred and three of our ARM and HSCR patients (48%) had an intestinal stoma on admission with an associated complication rate of 25% and about one out of five needed a surgical revision. A sigmoid-divided colostomy, as recommended before a PSARP,[4] was found only in 41% of children with ARM. However, recent studies have reconsidered the risk of distal loop contamination associated with a loop colostomy.[43] Antegrade washout of the distal loop before PSARP was often uneasy due to inspissated meconium remnants not being removed during emergency diversion,[44] and residual fecal debris was an obstacle to an antegrade high-pressure colostogram to visualize a recto-urinary fistula. Colon diversion for HSCR patients carried a minor complication rate (15%), mainly related to prolapse or stenosis.

Emergency levelling diversion for a suspected HSCR at a primary health level was based only on acute obstruction, abdominal distention, and a history of long-lasting constipation without any radiological or histological evidence. In many cases, the stoma was closed after aganglionosis was excluded. Retained fecalomas in the excluded bowel were hard to remove later, making pull-through procedures more complex.[40] To reduce the incidence of misdiagnosis and mismanagement, visitors from pediatric surgical outreach should expand their role in establishing closer links with primary health facilities in their catchment areas and promoting distant consultation to enhance the referral network.[3]

Table 3. Distribution of 108 ARM cases according to international Classification (Krickenbeck's).

	Males 50	Females 58
Perineal fistula	10	4
Rectouretroperineal fistula	1	
Anal stenosis	1	
No fistula	1	
Recto urinary fistula		
- Bulbar	26	
- Prostatic	5	
- Vesica	5	
Vestibular fistula		47
Cloaca		3
Rectal atresia	1	2
H Type fistula		2

Constraints to corrective treatment and barriers to post-discharge monitoring and follow-up

The pediatric surgical workforce has expanded significantly in many SSA areas over recent decades.[45,46] Unfortunately, a profound imbalance exists between the capacity to care and the ability to afford it.[47,48] Access to healthcare is impaired for most children as the source of funding for diagnosis, treatment, and follow-up is generally from out-of-pocket payments by most low-income patient's families. In our experience, the failure to respect treatment and follow-up schedules was less due to cultural constraints and more to financial problems related to long-distance travelling from unconnected rural areas, hospital fees, and long-lasting hospital treatments interfering with parents' working activities. One of our hospitals was a charitable institution with reduced costs for children living in the district. However, the other two governmental institutions required some form of payment from most patients without a financial protection scheme. The benefits of short-term outreach missions are generally reflected in the quality of treatments rather than in health coverage and access to care.

If adequately trained, community health workers or nurses/midwives could provide early recognition, referral, and long-term follow-up of PCRD at rural and primary health levels. Investments in increasing the number of medical specialists in SSA do not extend to the full range of non-physician healthcare professionals. Their limited involvement and low wages lead to low motivation and subpar performance. More attention to these aspects in recruitment, training, and retention is needed to upgrade the standard of care.[49]

Our figures confirm that inadequate long-term follow-up remains a critical issue in treating PCRD in a low-resource context. Most published series[1,44,50] report figures on the number of procedures and short-term functional results but are somewhat elusive about the regularity and duration of patient attendance at the follow-up schedule. In a poor cultural background, the immediate relief from corrective surgery, associated with financial restraints to travel and access health services, leads families to underestimating the need and the value of periodic controls to prevent the two main long-term sequelae of PCRS surgery: constipation and incontinence. In SSA, the quality of specialist surgical services is growing faster than in establishing a network of accessible healthcare services.[51] Those patients living in poor, rural, underserved areas who are facing constipation or poor continence after a technically flawless PSARP or pull-through is done either by a local specialist or a short-term visiting surgical team, but who have been left without adequate post-discharge care support, are at risk of a life of

misery and exclusion. A diverting stoma was preferable to them for a better quality of life.[52] It is a great responsibility for outreach initiatives to be based on a long-term, local perspective of comprehensive care, seeking full integration with and the broadest possible involvement of local health resources. The mere focus of local and visiting surgeons on the progress of surgical skills, the number of procedures, and the short-term results risks leaving in the background that most African pediatric patients still lack the necessary follow-up for an effectively successful PCRD surgery.[1,20,50,53]

Context-aware adaptive responses

The management of PCRD in underserved areas of SSA faces unique challenges. Local and visiting surgeons must have adaptive capacities and sustainable solutions.

A high rate of stoma-related complications (from 20 to 70%) is reported strictly related to the surgeon's low exposure to pediatric surgery at a primary level of care.[54] Although loop colostomy, which is undoubtedly easier to create and close, has been considered for a long time unsuitable for patients needing a PSARP, more recent views consider both divided and loop colostomy acceptable for faecal diversion, with similar risks and benefits.[43,55] Although we generally prefer to create a divided colostomy in contexts with a higher risk of infection, poor nursing care, and low availability of stoma bags, we saved our patients the revision of a loop stoma by preferring a temporary purse-string closure of the distal stoma to prevent post-PSARP wound contamination.

The prevention of perineal wound infection has always been our primary concern due to the scarcity of nursing staff, poor hygienic conditions, a shortage of cleaning products, and the abundance of mucous discharge from the new perineal anal opening, which contribute to infection and dehiscence. Although our rate of sepsis and perineal wound dehiscence (9.2%) is within the range of other reports from SSA,[24] we modified the PSARP technique, leaving a temporary long, closed rectal stump for two weeks[6] to keep the infection rate low and to reduced hospital stay and the need for catheterisation. Our patients tolerated the procedure well, with no major discomfort. Cosmetic results were satisfactory, and there was no delay in starting the dilatation protocol.

Within the scope of reducing the number of admissions and hospital stays, as well as lowering the risks associated with colostomy closure, in a context where one-step procedures are impossible without frozen sections, we opted for a two-step pull-through for HSCR. The straightforward pull-through of the ganglionic stoma has a possible limitation in the risk of including a tract of a healthy bowel in the resection if the site for the levelling colostomy has been chosen without the help of frozen sections. For this reason, we usually take multiple specimens from the distal colon during the sigmoid levelling colostomy confection to examine later. This practice is generally ignored by surgeons confectioning a stoma at a primary health level. Nevertheless, we found a very short ganglionic tract included in the resected tract only in four of our patients.

A last challenge for PCRD surgery in under-resourced areas is long-term patient follow-up.[53] Our figures confirm the difficulties in maintaining contact and tracing patients with poor attendance at scheduled surgical programs or postoperative checks. Our adaptive strategy was based on the involvement of local health providers in achieving surgical skills through on-the-job training and coaching, as well as keeping patients' files updated, with special regard to the long-term outcomes of operated patients. At the same time, we helped maternal post-PSARP home care by supplying a dilators' set and training mothers on how to use them. Our nurses also demonstrated bowel management techniques to prevent post-PSARP chronic constipation and HSCR treatment through dietary modifications to mitigate the effects of poor continence. If money and distance hinder regular access to health services, relying on well-trained mothers is the only way to prevent long-term complications of colorectal surgery in children. To make sense, surgical outreach in a low-resource context must devote a significant portion of the visiting teams' work to educational targets.[56]

Conclusions

In the realm of surgical outreach, adaptive responses are essential for the successful management of PCRD in under-resourced contexts. Visitors must skillfully navigate the complex landscape of healthcare challenges, tailoring their strategies to meet the diverse needs of these children who are often stranded without adequate surgical care. Revision of streamlined procedures and protocols is needed to reduce hospital stays, shorten the intervals between crucial treatment steps and minimise complications, ensuring that every child receives timely care without compromising safety or the quality of outcomes. Follow-up length and quality can also be improved significantly if specific strategies are adopted. Collaboration with local healthcare staff is equally critical, without overwhelming them and defrauding them of their role by employing overly large outreach teams. Partnerships must value shared knowledge and experiences, empowering local caregivers and enhancing their capacity to provide a higher standard of care through sustainable models of clinical governance and better management of human, technological, and infrastructural resources. Embracing these strategies can significantly elevate the standard of healthcare development cooperation aid.

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