Are end colostomies always contraindicated in anorectal malformations?

G Brisighelli,[1,2] MD (FC Paeds Surg); **A P Theron**,[1,3] MMed;
C Westgarth-Taylor,[2,4] MB ChB, FC (Paeds Surg); **E M Mapunda**,[1,3] MBBS, MMed (Paed Surg)

[1] *Division of Paediatric Surgery, Department of Surgery, Faculty of Health Sciences, University of the Witwatersrand, Johannesburg, South Africa*
[2] *Johannesburg Paediatric Colorectal Clinic, Department of Paediatric Surgery, Chris Hani Baragwanath Academic Hospital, Johannesburg, South Africa*
[3] *Department of Paediatric Surgery, Charlotte Maxeke Johannesburg Academic Hospital, Johannesburg, South Africa*
[4] *Center for Colorectal and Pelvic Reconstruction, Nationwide Children's Hospital, Columbus, Ohio, United States*

Corresponding author: *A Theron (andre.theron@gmail.com)*

Background. In our clinical setting, a three-staged approach is consistently employed to manage patients with anorectal malformations (ARMs).
Objective. To evaluate the safety and feasibility of end-colostomies, in particular subtypes of ARMs.
Methods. The medical records of babies born with an ARM treated between 1 January 2017 and 31 December 2022 were reviewed. Information regarding the type of ARM, type of colostomy, complications during colostomy formation, posterior sagittal anorectoplasty (PSARP) and colostomy closure was recorded.
Results. Ultimately, 194 patients were included. Of those, 137 patients had divided colostomies with distal mucus fistulas (DC) and 57 had end-colostomies (EC). Ninety-seven patients (50%) had perineal and vestibular fistulas, including 40 (41%) patients with DC and 57 (59%) with EC. For post-colostomy formation, eight (20%) complications were recorded in the DC group and nine (16%) in the EC group (p=0.78). Wound sepsis presented in six patients with DC and four with EC (p=0.3). Nineteen percent (16/85) of patients who had PSARP developed complications, including 4/37 (11%) with DC and 12/48 (25%) with EC (p=0.16). Of the 72 patients who underwent stoma reversal, six (8%) had complications post-operatively: three in the DC group and three in the EC group.
Conclusion. For patients with rectoperineal and vestibular fistulas, where divided colostomies are not warranted, end colostomies provide a safe alternative.

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Anorectal malformations (ARMs) are among the most prevalent congenital malformations often requiring a multitude of operations and tailored medical and surgical management. While some types of ARMs are amenable to a primary posterior sagittal anorectoplasty (PSARP), others necessitate a three-staged approach consisting of formation of a colostomy, deferred definitive repair by PSARP and subsequent closure of the colostomy. [1,2]

A devastating complication following a PSARP is wound sepsis, which can compromise nerves and muscles, resulting in poorer outcomes in bowel control. Consequently, a three-staged approach may be justified, particularly in low-middle-income countries (LMICs) and Sub-Saharan regions where the risk of wound sepsis is elevated.[3] In this approach, colostomy formation plays a crucial role by decompressing the obstructed bowel, preventing faecal contamination of the urinary tract in the presence of a recto-urinary fistula and protecting the future perineal reconstruction.[3,4] The ideal type of stoma for children with ARMs remains a subject of controversy.[1] The two most described colostomies are the divided descending colostomy with a distal mucus fistula and the loop colostomy.[4] The divided descending colostomy is often preferred over the loop colostomy because of a lower incidence of prolapse and urinary tract infections.[2-4] End colostomies are generally contraindicated irrespective of the type of ARM. This is primarily because they limit the ability to perform an augmented pressure distal colostogram at a later stage.[1,2,5] We, however, believe that specific types of ARMs, mainly rectoperineal and recto-vestibular fistulas, could benefit from an end colostomy as the presence of a mucus fistula to decompress the defunctionalised bowel is not needed and performing an augmented pressure distal colostogram would not be necessary to delineate the anatomy before the PSARP.

Our study aimed to provide a comprehensive analysis of our experience with the use of end colostomies in patients with rectoperineal and vestibular fistulas. By examining the outcomes and complications associated with this approach, we aim to contribute valuable insights to the ongoing discussion on the optimal type of colostomy for children with ARMs.

Methods

The research project was approved by the Human Research Committee at the University of the Witwatersrand (ref. no. M200533).

Data were obtained by retrospectively reviewing medical records of patients diagnosed with an ARM and had a stoma fashioned between 1 January 2017 and 31 December 2022 at the two academic hospitals affiliated with the University of the Witwatersrand (Charlotte Maxeke Johannesburg Academic Hospital (CMJAH) and Chris Hani Baragwanath Academic Hospital (CHBAH)).

Both hospitals use an electronic database, from which we retrieve patient information. Information regarding the sex of the child, type of ARM, type of colostomy fashioned and complications (long and short term) associated with the fashioning of the colostomy were reviewed. Data regarding age at stoma formation were also recorded, along with information on the HIV status of both the mother and the child. Patients

were categorised as either HIV-exposed or HIV-unexposed.

The incidence of surgical complications at stoma formation, PSARP and stoma reversal was also analysed and compared between patients who underwent a divided colostomy and an end colostomy. A comparison of the incidence of surgical complications during the three stages was also performed including only patients with good-prognosis ARM (perineal and vestibular fistulas). This was done based on the assumption that poor-prognosis ARMs are associated with longer operating durations and larger wounds, thereby posing a higher risk of surgical complications compared with good-prognosis ARMs.

Data were collected with Microsoft Excel and statistical analyses were performed with GraphPad (GraphPad Software, USA) using the Fisher's exact test (for dichotomous variables) and Student's *t*-test (for continuous variables). A *p*-value <0.05 was considered statistically significant.

Patients were excluded if the initial colostomy was performed at another institution, if they passed away or were placed under palliative care owing to the severity of associated anomalies before the stoma was fashioned, if they underwent a primary PSARP without an initial colostomy and if they presented with a rare form of ARM (e.g., cloacal exstrophy, covered cloacal exstrophy, posterior cloaca or aphallia spectrum). Patients with incomplete records as well as those who demised within two weeks of stoma fashioning owing to complications associated with the severe concomitant anomalies (cardiac, rena or oesophageal atresia) were also excluded.

Surgical technique

The surgical procedure commences with the child placed in a supine position on the operating table. A Foley catheter or a nasogastric tube is inserted into the fistula and advanced approximately 20 cm (Fig. 1A). Following draping, a small circular incision measuring 2 cm in diameter is made in the left iliac fossa and excision of the skin and subcutaneous tissue in the area is performed (Fig. 1B). Subsequently, an oblique incision is meticulously created in the external oblique muscle, facilitating the delivery of the colon. During this step, careful palpation is employed to locate the Foley catheter, ensuring accurate identification and orientation of the proximal and distal colon. At the specific level typically designated for divided colostomy, which is positioned just distal to the junction of the descending and sigmoid colon, the bowel is divided. The distal end of the bowel is meticulously oversewn, and to optimise subsequent stoma reversal, it is hitched to the adjacent peritoneum. Following this, the proximal end of the bowel is exteriorised and secured to the sheath. Maturation of the proximal end to the skin is then accomplished, thereby completing the colostomy formation (Fig. 1C).

Results

In the time frame considered, 236 consecutive children born with an ARM were treated in the two hospitals (CMJAH: *n*=104, 44% and CHBAH: *n*=132, 56%). Forty-two patients (18%) were excluded, 21 (50%) underwent a primary PSARP, seven (17%) with severe associated anomalies and died before any surgical intervention, five (11%) with a colostomy opened elsewhere, four (10%) with a rare type of ARM, four (10%) who died within 2 weeks of stoma fashioning (owing to severe associated anomalies) and one (2%) with missing data. Of the 21 excluded patients who underwent a primary PSARP, 14 were male (*n*=10 with rectoperineal fistulas and *n*=4 with unknown ARM type) and seven were female (*n*=5 with rectoperineal and *n*=2 with recto-vestibular fistulas). Two patients who underwent a primary PSARP developed wound sepsis and required a protective end colostomy. One of the two patients required a redo PSARP before stoma reversal.

In total, 194 patients were included (*n*=111, 57% male). The median age at stoma formation was 3 days (range 0 - 2 667; interquartile range 16). The different types of ARMs are presented in Table 1.

Forty-five (23%) patients were HIV-exposed, 95 (49%) were unexposed and the status of 54 (28%) patients was unknown.

Of the included patients, 137 (71%) had divided colostomies with distal mucus fistulas (13 of them required a formal laparotomy with a divided colostomy owing to severe abdominal distension, presence of a hydrocolpos, intraoperative findings of malrotation, or bowel perforation), and 57 (29%) end colostomies (four of them required a midline laparotomy: two because of severely distended abdomens, one because of a pneumoperitoneum, and one owing to intraoperative findings of malrotation). The median age at stoma formation was 2 days (range 0 - 2 667; IQR 3) for divided colostomies and 26 days (range 0 - 1 310; IQR 150) for end colostomies (*p*=0.03).

The type of colostomy fashioned according to the ARM type is summarised in Table 2.

Thirty-three of 137 (24%) patients with a divided colostomy were HIV-exposed compared with 12/57 (21%) in the end colostomy group (*p*=0.8).

Overall, 55 patients (28%) developed stoma complications: 46/137 (34%) with divided stomas and 9/57 (16%) with end colostomies (*p*=0.014). Median age at stoma formation was two days (range 0 - 491; interquartile range 3) for the patients who developed complications and 4 days (range 0 - 2 667; IQR 23) for patients who did not develop complications (*p*=0.13).

Regarding the type of complications, 35 (18%) patients developed wound sepsis (25 treated conservatively), five had a retracted proximal stoma needing revision, five developed an adhesive small bowel obstruction, five had a retracted/stenosed mucus fistula (with four needing a revision), two had an intraoperative finding of malrotation and required a formal laparotomy and a Ladd's procedure, one had a volvulus with a small bowel perforation, one had inverted stomas and one had a prolapsed mucus fistula that was managed conservatively. Table 3 summarises the types of complications according to the type of stoma.

Seventeen of the 45 (38%) HIV-exposed patients developed complications at stoma opening compared with 25/94 (27%) HIV-unexposed patients (*p*=0.23).

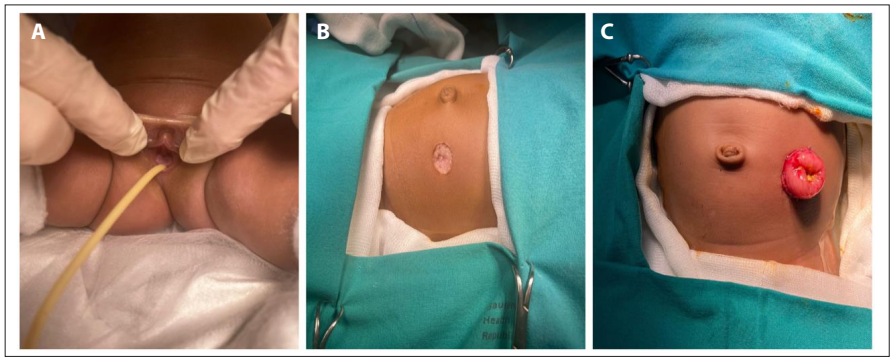

Fig. 1A. Insertion of urinary catheter into recto-vestibular fistula. Fig. 1B. Abdominal skin and subcutaneous incision. Fig. 1C. Formation of colostomy.

Of the 194 patients, 157 (81%) had a PSARP performed, 10 (5%) died before PSARP owing to associated anomalies (mainly cardiac and tracheoesophageal) and 27 (14%) were awaiting PSARP.

In total, 97 (50%) patients had recto-perineal or recto-vestibular fistulas, with 57 (58%) receiving an end colostomy and 40 (42%) a divided colostomy. At the stoma opening, 17 complications were recorded: 9/57 (16%) in the end stoma group and 8/40 (20%) in the divided stoma group (p=0.78). Two patients in each group were HIV-exposed (p=1.00). Wound sepsis was the most frequent complication, with 4/57 (7%) patients developing wound sepsis in the end stoma group and 6/40 (15%) in the divided stoma group (p=0.31). All wound sepsis was treated conservatively. Table 4 summarises the types of complications according to the type of stoma in patients with rectoperineal and vestibular fistulas.

Of the 97 patients with rectoperineal and vestibular fistulas, a PSARP was performed in 85 patients and is still pending in nine patients (seven in the end stoma group and two in the divided stoma group) and three patients died before PSARP owing to associated anomalies. In total, 16 (19%) complications were recorded after PSARP: 12/48 (25%) patients with end colostomies compared with 4/37 (11%) with divided colostomies (p=0.16). The most common complication was stricture of the anoplasty which occurred in eight patients: six (13%) in the end stoma group and two (5%) in the divided stoma group (p=0.28). Wound sepsis occurred in eight patients: 6 (13%) in the end stoma group and two (5%) in the divided stoma group (p=0.46). Two patients with an end stoma developed a complete stricture of the PSARP, secondary to wound sepsis, requiring a redo PSARP. They did not

Table 1. Type of anorectal malformations (ARMs) according to sex in the study population

Male, *n* (%)	**111 (57)**
ARM (recto-urethral bulbar)	31 (28)
ARM (recto-perineal)	30 (27)
ARM (unknown)	16 (14)
ARM (imperforate without fistula)	16 (14)
ARM (recto-urethral prostatic)	10 (9)
ARM (bladder neck)	7 (7)
ARM (rectal atresia)	1 (1)
Female, *n* (%)	**83 (43)**
ARM (recto-vestibular)	49 (59)
ARM (recto-perineal)	16 (19)
ARM (cloaca)	12 (14)
ARM (imperforate without fistula)	3 (4)
ARM (recto-vaginal)	2 (2)
ARM (anal stenosis)	1 (1)
Total	**194 (100)**

ARM = anorectal malformations.

Table 2. Type of stoma fashioned according to the type of ARM

Divided colostomy, *n* (%)	**137 (71)**
ARM (recto-urethral bulbar)	31 (16)
ARM (recto-perineal)	20 (10)
ARM (imperforate without fistula)	19 (10)
ARM (recto-vestibular)	19 (10)
ARM (unknown)	16 (8)
ARM (cloaca)	12 (6)
ARM (recto-urethral prostatic)	10 (5)
ARM (recto-bladder neck)	7 (4)
ARM (recto-vaginal)	1 (1)
ARM (rectal atresia)	1 (1)
ARM (anal stenosis)	1 (1)
End colostomy, *n* (%)	**57 (29)**
ARM (recto-vestibular)	30 (15)
ARM (recto-perineal)	26 (13)
ARM (recto-vaginal)	1 (1)

ARM = anorectal malformations.

Table 3. Complications according to type of stoma in all patients with anorectal malformations

Type of complication	DC, *n/N* (%)	EC, *n/N* (%)	*p*-value
Wound sepsis (conservative treatment)	21/46 (46)	4/9 (45)	1.00
Wound sepsis (requiring surgical intervention)	10/46 (23)	0/9 (0)	0.18
Retracted mucus fistula (n=4 requiring revision)	5/46 (12)	0/9 (0)	0.60
Retracted proximal stoma requiring redo	4/46 (9)	1/9 (11)	1.00
Adhesive small bowel obstruction	2/46 (4)	3/9 (33)	0.02
Prolapsed mucus fistula	1/46 (2)	0/9 (0)	1.00
Inverted stoma	1/46 (2)	0/9 (0)	1.00
Intraoperative finding of malrotation	1/46 (2)	1/9 (11)	1.00
Volvulus on adhesions (demised)	1/46 (2)	0/9 (0)	1.00

DC = divided colostomy; EC = end colostomy.

Table 4. Type of complications according to type of colostomy in patients with recto-perineal and recto-vestibular fistulas

Type of complication	DC (N=40), *n/N* (%)	EC (N=57), *n/N* (%)	*p*-value
Wound sepsis (conservative treatment)	6/40 (15)	4/57 (7)	0.31
Adhesive small bowel obstruction	1/40 (3)	3/57 (5)	0.64
Retracted proximal stoma requiring redo	1/40 (3)	1/57 (2)	1.00
Intraoperative finding of malrotation	0/40 (0)	1/57 (2)	1.00

DC = divided colostomy; EC = end colostomy.

require a stoma revision to perform an augmented pressure distal colostogram before the redo PSARP.

A stoma closure was performed in 72 patients: 36 with end stomas and 35 with divided stomas. Eleven stoma closures were still pending and two patients died between PSARP and stoma reversal (one because of acute gastroenteritis and one owing to cardiac issues). A total of six complications (8%) were recorded after stoma reversal: three in the end colostomy group (two wound sepsis and one anastomotic breakdown) and three in the divided colostomy group (wound sepsis, anastomotic stricture, and anastomotic breakdown) (p=1.00).

Discussion

ARMs are a highly prevalent congenital anomaly and the most frequent cause of neonatal intestinal obstruction in Africa.[6] In 2010, in the greater Johannesburg area, the birth prevalence of ARMs was 1 in 3 989 births.[6] Nearly half of the patients with ARMs treated at our institution experience a delayed diagnosis and over 20% of children have a history of HIV exposure.[7,8] Both factors elevate the risk of complications among children, especially sepsis. Consequently, in our setting, a three-stage approach is preferred for managing all ARMs, including malformations with favourable prognoses such as rectoperineal and vestibular fistulas.[3,8-10]

The divided descending colostomy with distal mucus fistula is the preferred colostomy technique for patients with ARMs.[2,11] This technique effectively separates the proximal and distal stomas, thereby reducing the risk of prolapse and faecal contamination of the distal tract, consequently lowering the incidence of urinary tract infections in patients with recto-urinary fistulas.[1] Additionally, it allows the performance of an augmented pressure distal colostogram to delineate the anatomy of the ARM before the anorectal reconstruction is performed.[2] Owing to these reasons, loop colostomies and end colostomies are contraindicated in patients with ARMs.[1,2,11] In most centres, especially in high-income countries, a primary PSARP (or a delayed primary PSARP) would be the procedure of choice in patients with rectoperineal and vestibular fistulas. However, a primary PSARP is not always feasible in resource-limited settings owing to resource constraints. Consequently, even patients with good prognosis ARMs also undergo a three-stage approach.[3] Our experience corroborates findings from previous studies with less than 10% of all ARM patients undergoing a primary PSARP and most ARMs with good prognoses also receiving a stoma.[3] However, patients with rectoperineal and vestibular fistulas do not need a mucus fistula. They easily decompress from stool and mucus via the perineal opening and do not need an augmented pressure distal colostogram. The distal colostogram is usually discouraged in these patients as it may create the impression of a long, narrow fistula because of the inability to apply enough pressure, allowing the contrast to drain outside. Consequently, at our institution, patients with rectoperineal and vestibular fistulas, who exhibit a visible opening that allows appropriate decompression, undergo end colostomies. A divided stoma with a distal mucus fistula is still indicated if there is no visible perineal orifice. In our study, 42% of patients with recto-perineal or vestibular fistulas still received a divided stoma.

The median age at stoma formation was significantly higher in patients who underwent end colostomies compared with divided stomas. This is because only patients with perineal and vestibular fistulas received end colostomies. In these patients, the fistula allows for stool to decompress and therefore the malformation is sometimes diagnosed late or the colostomy can be selectively opened.

In the literature, the reported incidence of stoma-related morbidity ranges from 3 - 81%, with a mean complication rate of 45% for divided stomas.[12] Stoma prolapse, urinary tract infections, wound sepsis, stoma retraction and inverted stomas are the most frequently reported complications.[12,13] In LMICs, complication rates after stoma formation are higher compared with high-income countries (HIC), with peristomal excoriation and wound sepsis being the most frequently reported complications.[14] This is likely attributed to multiple factors, with delayed diagnosis, high HIV exposure, malnutrition, lack of trained stoma nurses and poverty being the most relevant ones.[8,15]

In our patient series, stoma complications occurred in 28% of cases, which is comparatively lower than the rates reported in the literature, particularly in LMICs.[12-14] Age at stoma formation did not correlate with an increased risk of complications. The most frequently observed complication was wound sepsis, even though most patients only required conservative treatment for this problem. In our series, the incidence of wound sepsis was higher in ARM patients with divided stomas compared with patients with end stomas, although this was not statistically significant. Moreover, none of the patients with an end stoma required surgical intervention for wound sepsis compared with 10 patients with divided stomas. We postulate that the presence of a suture line between the proximal and distal stomas contributes to the augmented risk of wound sepsis associated with divided stomas. Other factors, such as compromised nutritional status, HIV exposure and limited availability of nursing care may further heighten the vulnerability of these children to wound breakdown, thus favouring the use of end stomas as a preventive measure.[9,10,15-17] Adhesive small bowel obstruction was significantly more frequent in patients with end stomas compared with those with divided stomas. The exact reason for this finding remains uncertain, despite indications that decreased manipulation of bowel loops during end stoma creation could suggest a reduced risk of adhesion formation. However, it is plausible that the use of a surgical stapler to fashion the end colostomy could have increased the risk of inflammatory reaction and therefore adhesions if the stapler line was left in the abdomen.[18,19] This is, however, just a speculation as surgical notes are not always accurate and the use of a gastrointestinal stapler at stoma fashioning might not be documented. Based on these observations, we advise against the use of staplers to fashion colostomies in children with ARMs.

In addition to examining complications related to stoma formation, we considered it crucial to evaluate potential differences in the incidence of postoperative complications after PSARP and stoma closure. To mitigate the inherent bias associated with considering PSARP-related complications in all types of ARMs, we limited our analysis to patients with favourable prognoses, specifically rectoperineal and vestibular fistulas. Within this specific patient cohort, at stoma formation, the incidence of all complications is comparable between the divided stoma and end stoma groups, with wound sepsis remaining the most common complication. Nevertheless, no surgical intervention was required for wound sepsis in either group.

During PSARP, we observed an increased risk of wound sepsis in patients with end stomas compared with patients with divided stomas; this was, however, not statistically significant and most patients only required conservative treatment. We are unsure of the reason for the increased wound sepsis rate in the end stoma group because at our institution we do not routinely irrigate the mucus fistula before the PSARP. However, it is possible that in patients with end stomas, the defunctionalised bowel was not decompressed and washed out in the theatre during stoma creation, as is typically done in patients undergoing a divided stoma procedure. Therefore, we recommend, at stoma fashioning, to thoroughly wash out the

defunctionalised bowel for all patients regardless of the type of stoma that is being fashioned.

At stoma reversal, we did not observe statistically significant differences in complications among patients with end stomas v. those with divided stomas. However, the size of the wound was smaller in patients with end colostomies, thus possibly allowing for better cosmetic results.

Conclusions

In our clinical setting, we advocate for a three-staged approach as the preferred management strategy for all patients presenting with anorectal malformations, even those with favourable prognoses. Specifically, in the case of rectoperineal and vestibular fistulas, the requirement for a divided stoma is obviated owing to the inherent ability of the fistula to facilitate mucus drainage. Additionally, the performance of an augmented pressure distal colostogram, typically employed to provide further anatomical delineation, is not necessary in these cases. Drawing from our clinical experience, we conclude that end colostomies should offer notable advantages for these patients, as they do not appear to be associated with increased incidences of complications after stoma formation, posterior sagittal anorectoplasty or stoma closure procedures.

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