Original Article

Access this article online
Website: www.afrjpaedsurg.org
DOI: 10.4103/0189-6725.78663
Quick Response Code:

Management of anorectal malformation: Changing trend over two decades in Zaria, Nigeria

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ABSTRACT

Background: Anorectal malformation is a common congenital defect and its management has evolved over the years. This is a review of the trend in the management of this condition in a major paediatric surgical centre in Nigeria over two decades. **Materials and Methods:** A retrospective analysis of 295 patients with anorectal malformations managed from January 1988 to December 2007 was carried out. **Results:** There were 188 boys and 107 girls aged 1 day–9 years (median 8 years) at presentation. There were 73 (54.5%) and 106 (65.8%) emergency operations in groups A and B, respectively. There were 61 (45.5%) and 55 (34.2%) elective operations in groups A and B, respectively. Regarding treatment, in group A, patients requiring colostomy had transverse loop colostomy, while in group B, sigmoid (usually divided) colostomy was preferred. The definitive surgery done during the two periods were: group A: cutback anoplasty 29 (47.5%), anal transplant 5 (8.2%), sacroabdominoperineal pullthrough (Stephen's operation) 6 (9.5%) and others 21 (34.4%). In group B, posterior sagittal anorectoplasty (PSARP) 46 (83.7%), anal transplant 1 (1.8%), posterior sagittal anorectovaginourethroplasty (PSARVUP) 2 (3.6%) and anal dilatation 6 (10.9%) were done. Early colostomy-related complication rates were similar in the two groups ($P > 0.05$). The overall late complication rate was 65.5% in group A and 16.4% in group B ($P < 0.05$). The mortality was 25 (18.6%) in group A compared to 17 (10.6%) in group B ($P < 0.05$). **Conclusion:** There have been significant changes in the management of anorectal malformations in this centre in the last two decades, resulting in improved outcomes.

Key words: Anorectal malformation, evaluation, management, outcome

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INTRODUCTION

Anorectal malformation is a common cause of intestinal obstruction in children. The incidence is 1:4000–5000 live births worldwide, but this could be more.[1]

The classification and management of this condition has evolved over the years.[2-6] Initially, these patients were allowed to die without treatment.[2] Now, all patients with this condition can be appropriately treated. The surgical evolution of the treatment for anorectal malformation can be seen in procedures ranging from Stephen, Rehbein, Romualdi, Kiesewetter to Pena.[7]

This report is intended to establish the trend and experience in the management of anorectal malformation over two separate decades in a major tertiary centre in Nigeria.

MATERIALS AND METHODS

This is a retrospective review of medical records, operation registers and operation notes of patients managed for anorectal malformation at Ahmadu Bello University Teaching Hospital, Zaria, Nigeria, from January 1988 to December 2007. Demographic data, type of anomaly, mode of presentation, evaluation, treatment and outcome of treatment were obtained.

The patients were divided into two groups. Group A patients were managed from January 1988 to December 1997 and group B patients were managed from January 1998 to December 2007. Evaluation of early complications was based on colostomy-related complications among 54 patients in group A and 78 patients in group B, for whom adequate early complication records were available at the time of this study. Due to lack of reliable follow-up records, late complications in group A were based on patients whose complications required a procedure under general anaesthetic and operative records were used for this.

Data obtained were analysed using SPSS version 11.0. The χ^2 test was used to test for level of statistical significance. Level of statistical significance was set at 0.05.

RESULTS

There were a total 295 patients (188 boys and 107 girls). The median age at first surgery was 8 days (range 1 day–9 years). In group A, there were 134 patients (79 boys and 55 girls) of median age 8 days (range 1 day–13 years), and in group B 161 patients (109 boys and 52 girls) of median age 8 days (range 1 day–12 years) were studied. There were 73 and 106 emergency operations in groups A and B, respectively. The elective operations were 61 and 55 in groups A and B, respectively.

In group A, the main investigations for evaluation were a lateral invertogram and a sacral X-ray. In group B, the investigations were a lateral cross table X-ray, abdominal ultrasonography, sacral X-ray, echocardiography and a distal colostogram.

There has been a shift in diagnostic nomenclature in line with the changing international nomenclature. Terminology of imperforate anus used in group A was replaced by anorectal malformation in group B [Table 1].

In group A, 69 patients requiring colostomy had a transverse loop colostomy, while in group B 96 patients requiring colostomy had a divided sigmoid colostomy. In group A, 7 patients with rectovestibular fistula were given a colostomy before the definitive surgery, while in group B 14 patients with rectovestibular fistula were managed without a colostomy.

The definitive surgery done during the two periods were 61 and 55 for groups A and B, respectively. For group A, the surgeries were cutback anoplasty 29 (47.5%), anal transplant 5 (8.2%), sacroabdominoperineal pullthrough (Stephen's operation) 6 (9.8%), anal dilatation 11 (18.0%), endorectal pullthrough 6 (9.8%), cruciate incision 2 (3.3%) and excision of fistula 2 (3.3%). In group B, the operations were posterior sagittal anorectoplasty (PSARP) 46 (83.7%), anal transplant in 1 (1.8%), posterior sagittal anorectovaginourethroplasty (PSARVUP) 2 (3.6%) and anal dilatation 6 (10.9%) [Table 2].

Early complications were colostomy related [Table 3]. The early complication rate was 16.7% (9 of 54) in group A and 17.9% (14 of 78) in group B. The difference in early complication rate between the two groups was not statistically significant ($P > 0.05$).

Follow up in group A was difficult to determine as reliable post discharge records were not available. Of note, however, is the fact that only 12 patients who had colostomy returned for definitive surgery. In group B, 45 patients were followed up for 2 months–8 years (median 2 years).

In group A, late complications following definitive treatment were: faecal incontinence 1 (1.6%), anal stenosis 30 (49.2%) and rectal retraction 9 (14.7%); and for group B the complications were anal stenosis 5 (9.1%), mucosal prolapse 3 (5.5%) and faecal soilage 1 (1.8%) [Table 4]. The overall late complication rate in group A was 65.6% (40 of 61) and for group B it was

Table 1: Diagnosis for groups A and B

Diagnosis	Group A (%)	Group B (%)
Anorectal malformation	17 (12.7)	117 (72.7)
Imperforate anus	48 (35.8)	—
Anorectal agenesis	19 (14.2)	—
Anofourchette fistula	19 (14.2)	—
Anovestibular fistula	1 (0.7)	—
Perineal fistula	—	16 (9.9)
Persistent cloaca	—	5 (3.1)
Others	30 (22.4)	23 (14.3)
Total	134 (100)	161 (100)

Table 2: Definitive surgery for Groups A and B

Operation	Group A (%)	Group B (%)
Cutback anoplasty	29 (42.6)	—
Anal transplant	5 (8.2)	1 (1.8)
Sacroabdominoperineal pullthrough	6 (9.8)	—
PSARP	—	46 (83.7)
PSARVUP	—	2 (3.6)
Anal dilatation	11 (18.0)	6 (10.9)
Endorectal pullthrough	6 (9.8)	—
Cruciate incision	2 (3.3)	—
Excision of fistula	2 (3.3)	—
Total	61 (100)	55 (100)

Table 3: Early colostomy-related complications for groups A and B

Complication	Group A (%) n = 54	Group B (%) n = 78
Surgical site infection	—	6
Stoma separation and bowel evisceration	5	2
Stoma necrosis	—	3
Stoma retraction	2	—
Stoma stenosis	1	2
Peritonitis	1	—
Stoma bleeding	—	1
Total	9 (16.7)	14 (17.9)

Table 4: Late complications after definitive surgery for groups A and B

Complication	Group A (%)	Group B (%)
Faecal incontinence	1 (1.6)	—
Faecal soilage	—	1 (1.8)
Anal stenosis	30 (49.2)	5 (9.1)
Rectal retraction	9 (14.8)	—
Mucosal prolapse	—	3 (5.5)

Table 5: Causes of death for groups A and B

Cause	Group A (%)	Group B (%)
Anaesthesia	13 (9.7)	—
Overwhelming sepsis	8 (5.9)	11 (6.8)
Dyselectrolytaemia	4 (3.0)	1 (0.7)
Nonspecific	—	5 (3.1)
Total	25 (18.6)	17 (10.6)

16.4% (9 of 55). The difference in late complication rate in the two groups was statistically significant ($P < 0.05$).

In group A, there were 25 (18.6%) mortalities: anaesthetic deaths 13 (9.7%), overwhelming sepsis 8 (5.9%) and dyselectrolytaemia 4 (3.0%). In group B, there were 17 (10.6%) mortalities: overwhelming sepsis 11 (6.8%), dyselectrolytaemia 1 (0.7%) and nonspecific 5 (3.1%) [Table 5]. The mortality difference between the two groups was statistically significant ($P < 0.05$).

DISCUSSION

Anorectal malformation remains a common defect in paediatric surgical practice. There has been worldwide evolution in the management of this condition. The changing trends have been geared towards improving the outcome of patients managed with this condition. The most common cause of morbidity following the management of this condition is faecal incontinence. Therefore, one of the major challenges in the management has been to address this problem.

The embryological basis of the pathology has been extensively reviewed and has helped in the breakthroughs in the management of the disease.[8]

The classification of this disease is of paramount importance in order to properly group the malformations with a view to improve its management.[9,10] The earlier classifications such as Wingspread classification and the international classification did not fulfil a therapeutic objective. Pena's classification of the disease condition yielded appropriate therapeutic decisions and was therefore adopted during the second decade (group B). It is pertinent to note that the classification is still undergoing evolution. In a recent international conference on anorectal malformation held in Krickenbeck, Germany, new terminologies such as H-type malformations, rectal pouch and others have been introduced in the nomenclature.[6,11]

In our centre, the terminology of imperforate anus which was commonly used during the first decade, was replaced by anorectal malformation in the second decade.

In both the groups, most of the patients presented as newborns to the emergency department. This is because anorectal malformation commonly presents with intestinal obstruction which is an emergency. This picture was similar to another report from Nigeria.[12] However, patients in groups A and B frequently presented late with marked abdominal distension.

The lateral invertogram, which was a modality of investigation in group A, was replaced by the lateral cross table X-ray in group B. This lateral invertogram could be associated with aspiration pneumonitis or cerebral congestion when performing the procedure. The introduction of the lateral cross table X-ray was to avoid this risk.

Distal colostogram, abdominal ultrasound scan, as well as echocardiography were introduced in group B. The distal colostogram helps to outline the nature of rectourinary fistula as well as the level of blind rectal pouch, which are critical to the success of PSARP. The echocardiography and abdominal ultrasound scan are important in isolating associated cardiac and renal anomalies. These anomalies, when present, could influence the outcome of this condition adversely. Both abdominal ultrasound scan and echocardiography were not used in group A because they were not readily available in our centre at that time.

Another changing trend was in the management of anorectal malformation with rectovestibular fistula. In the later part of the second decade (group B), patients with this condition were managed without a preliminary colostomy, with good outcome. This is different from group A where an initial colostomy was done before the definitive procedure. It was noted that proper bowel preparation before definitive surgery was adequate for those who had definitive surgery without a colostomy. One stage treatment of vestibular fistula has also been advocated by others.[13,14,15]

In terms of early complications following initial treatment, colostomy-related complication rates were high but similar in the two groups. The overall late complication rate was significantly higher in the first decade of the study. However, poor follow up in the first decade makes it difficult to obtain a clear picture of late complication rates following definitive treatment. Although anal stenosis occurred in both the decades, stenosis in group B was not severe and the patients responded well to outpatient dilatation. This may be largely because there was a well-planned dilatation schedule after PSARP in group B. Mucosal prolapse was also noticed following PSARP, as in another report.[16] Those with major mucosal prolapse had mucosal excision and repair to address the prolapse. One patient developed faecal soilage after PSARP in group B, due to lack of toilet training. The faecal soilage, however, abated after proper toilet training.

The overall outcome from this study showed that there was some improvement during the second decade (group B) in management, when compared to the first decade (group A). This was exemplified by improvement in mortality rate and the nature of morbidity between the two groups. This has been observed in other reports also.[17]

CONCLUSION

There has been a significant change in the management of anorectal malformation with improved outcome in our centre. This has been due to better understanding of the pathological condition and refinements in the surgical treatment.

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Cite this article as: Lukong CS, Ameh EA, Mshelbwala PM, Jabo BA, Gomna A, Akiniyi OT, Nmadu PT. Management of anorectal malformation: Changing trend over two decades in Zaria, Nigeria. Afr J Paediatr Surg 2011;8:19-22.

Source of Support: Nil, **Conflict of Interest:** None declared.