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Preventing posterior sagittal anoplasty *'cripples'* in areas with limited medical resources: A few modifications to surgical approach in anorectal malformations

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ABSTRACT

Background: Anorectal malformations (ARM) are the most common neonatal emergencies in Sub-Saharan Africa countries. Late presentation, lack of pediatric facilities and trained paediatric surgeons influence the outcome of these patients. This study reports a 5-year of experience in the management of ARM at the Orotta Referral Hospital in Asmara (Eritrea) and proposes some modified surgical approaches to minimize the risk of complications and the length of hospital stay. **Materials and Methods:** We reviewed the records of 38 patients with ARM observed between September 2006 and April 2011. Since 2009 a modification of original posterior sagittal anorectoplasty (PSARP) was introduced, consisting in a long rectal stump (3 cms) closed and left at the perineal level, to be trimmed after two weeks. This avoided mucous spillage on the wound and prevented contamination. Post-operative course and outcome were evalued in the two group of patients divided according the type of surgical technique (Group A: Standard PSARP; Group B: Modified PSARP). **Results:** There were 21 boys and 17 girls aged 4 days to 9 years (median age 182 days). Of the 38 patients, 2 infants died before surgery and 3 refused preliminary colostomy. Previously confectioned colostomies often required revision or redoing due to severe prolapse or malposition. When possible, primary sigmoid colostomy was performed. There were 15 patients in Group A and 18 in Group B. Wound infection or disruption were recorded in 7 cases (46%) in Group A and in 2 (11%) in Group B. Late complications were related to anal stenosis, which required long term dilatations. Three cases needed a PSARP redo (2 in Group A, 1 Group B). **Conclusions:** We believe that our simple modification of original PSARP technique could be of help lowering post-operative complications rate and reducing hospital stay. Family compliance is mandatory for long-term surgical success. A relevant time must be spent in training to stoma care and post-operative anal dilatation.

Key words: Anorectal malformation, colostomy, complications, modified posterior sagittal anorectoplasty

INTRODUCTION

Anorectal malformations (ARM) are the most commonly reported cause of congenital intestinal obstruction in Sub-Saharan Africa with a higher estimated incidence than in other parts of the world.[1] This figures could be influenced by a higher hidden mortality among other gastrointestinal (GI) anomalies, easily missed in under resourced contests. As far as ARMs are concerned, males can sometimes survive enough for a life-saving colostomy; females evacuate through a vaginal fistula and diagnosis may be delayed. Colostomy is preliminary to reconstructive surgery and may be challenging to perform whenever even minimal medical facilities are lacking and strong resistance is encountered by parents. When poorly performed, stomas may be an added factor of morbidity and mortality. Only a properly done posterior sagittal anorectoplasty (PSARP) a meticulous post-operative management and a strict follow-up protocol can give these patients a good quality of life avoiding failures and complications. A five-year experience operating ARM at the Orotta Referral Hospital in Asmara, Eritrea is reported. The risk of post-operative complications due to scarce nursing resources and reduced hospitalization time are the main limiting factors for surgical success and long-term functional results. The aim of this work was to find solutions to these problems and to suggest some "tricks of the trade" to limit failures, simplify post-operative care and reduce nursing workload.

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MATERIALS AND METHODS

A retrospective study was carried out on 38 patients admitted for ARM at Orotta Hospital in Asmara, Eritrea, between September 2006 and April 2011. During that same time, a paediatric surgical assistance and training partnership program was developed in agreement between the Eritrean Ministry of Health and the San Camillo-Forlanini Hospital of Rome, Italy. Along three missions per year, each lasting 3 weeks, teams of two surgeons, one anaesthetist, and one paediatric nurse worked in full collaboration with local medical and surgical staff to improve the local standard of paediatric surgical care. Data about sex, age, type of ARM, associated anomalies, presence and location of a stoma, were extracted. Surgical choices, post-operative course and complications were analysed. Follow-up data, when available, were also reviewed. Our protocol included a thorough study of patients to assess general, clinical, and nutritional status and the existence of additional congenital anomalies. Pre-existing stomas were examined in order to evaluate location and function. Colostomy was revised or re-confectioned in all cases where it was severely prolapsed or failed to exclude distal bowel. A primary diverting sigmoid colostomy was performed, when possible, in all other patients. Unsuccessful attempts of perineal surgery made by other non-paediatric surgical teams were considered for revision after colostomy re-confectioning when necessary. Surgical strategies were individualized according to existing conditions and the social and cultural state of families. Posterior sagittal anorectoplasty (PSARP) according to the original Peña technique[2] was the procedure of choice after meticulous bowel preparation. Dilatations of the new anus, using a set of disposable Hegar bougies, were started two weeks after. Parents were trained to carry on with this maneuverer, after returning to their communities, until proper anal size for age was achieved. Colostomy was usually closed after 6-8 weeks if anoplasty site appeared completely healed and of proper caliber. Anal dilatations protocol extended for one year after PSARP and parents were recommended to come to our outpatient clinic every three months. Beginning in 2009 a technical modification of original PSARP was introduced due the need to protect perineal wound from anal mucous discharge. This was a major problem in an area affected by a shortage of nursing care and cleaning devices. Modification consisted of leaving a long rectal stump (3 cms) anchored by eight vicryl stitches to the perineal skin at the anal opening and closed by a double purse-string suture [Figure 1]. This helped to keep the perineal area clean and to prevent local infections and suture disruption. Discharge from hospital was usually planned 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 [Figures 2a and 2b]. The impact of this technical modification on post-operative course and



Figure 1: The rectal stump closed and left at the perineal level



Figure 2a: Trimming of the rectal stump performed two weeks after the surgery



Figure 2b: The cosmetic outcome

final results were evaluated dividing our patients in two groups (A and B) according to the type of anorectoplasty adopted (original or modified PSARP). Incidence of wound infections or dehiscence was compared. Late problems like anal stenosis or prolapse, constipation, soiling, poor continence were also evaluated. The data were analysed using GraphPad Instat® version 3.10 software. The Fisher's exact test was used for categorical variables. The *P* value equal to 0.05 or less was considered significant.

RESULTS

Thirty-eight patients were admitted for ARM: 21 boys and 17 girls (M: F: 1.2:1). The age at presentation ranged from 4 days to 9 years, with a median age 182 days. There were only two major associated anomalies (1 Fallot tetralogy, 1 Duodenal stenosis). Seventeen patients had a colostomy at the time of admission; four also had undergone previous perineal surgery at a peripheral hospital with unsatisfactory results. Whenever a colostomy was present, it was on the sigmoid in 11 and was prolapsed in 4 of them. Six patients had a transverse colostomy and 4 of them were prolapsed. Four stomas needed redoing. Characteristics of patients, management and outcome are summarized in Table 1. Only 33 of 38 patients could benefit from a complete surgical treatment. Two male infants, referred with a severely prolapsed transverse colostomy, died before surgery because of poor general conditions secondary to severe malnutrition. Three female patients were referred late for a previously undiagnosed recto-vaginal fistula and families refused preliminary colostomy before surgical correction of their anomaly. Comparing results of Group A and B, the number of perineal infection and wound dehiscence was significantly lower among cases that received a modified PSARP ($P = 0.0469$). Late complications were mainly related to anal stenosis secondary to wound infection or disruption. It generally resolved with bouginage, but required two PSARP redoing among Group A and one among Group B. Unfortunately only 10/15 and 12/18 patients in the two groups could benefit from enough follow-up lengthy to evaluate families' compliance with dilatations and middle term results.

DISCUSSION

ARM is a common congenital malformation with an incidence in developed countries of 1 in 5,000 live births. Among sub-Saharan African population, ARM represents 67% of neonatal emergency surgical procedures.[1,3] There are no reliable studies of incidence of ARM in Eritrea. Mortality related to ARM is still high in Africa (30.5%) because most patients are referred late for medical attention. Adejuyigbe *et al.*,[4] reports that only 15% are observed within 24 hours of life. Male infants present vomiting and progressive abdominal distension and are destined to die whenever colostomy is not performed on emergency basis. Limited hospital facilities, long distances, inadequate transport, dearth of surgeons trained to cope with these anomalies contribute all to maintain a high mortality rate.[5-7] Associated anomalies influencing mortality among ARM are usually reported.[3] The apparently low incidence of associated anomalies in our series could be possibly related to a mechanism of selection, which prevented referral of most severe cases from distant areas, in time to be assisted. Local surgeons provide ARM a generally inadequate standard of care with a consequent high rate of complications. In our study, four cases had a failed attempt of perineal surgery with permanent damage to anal muscular complex and 6 out 17 emergency colostomies had been performed on the transverse colon. None of them excluded the distal loop,

Table 1: Comparison between original and modified posterior sagittal anorectoplasty: characteristics of patients, postoperative course and outcome

	Group A	Group B
No. of patients	20	18
Male/Female	11/9	10/8
Type of anomaly	Rectourethral fistula 9 Rectovesical fistula 1 Congenital anal stenosis 2 Rectovaginal fistula 2 Rectovestibular fistula 6	Rectourethral fistula 8 Rectoperineal fistula 1 Congenital colon pouch 1 Rectovestibular fistula **7** Cloacae 1
Colostomy present	8	9
Site	*4 Transverse / 4 Sigmoid*	*2 Transverse / 7 Sigmoid*
Prolapse	*4*	*4*
Colostomy Redo	2	2
Previous perineal surgery failed	3	1
Colostomy and PSARP refused	3	-
Dead before PSARP	2	-
No. of patients who received PSARP	Original PSARP 15	Modified PSARP 18
Wound infection/dehiscence	7	2
Lost during follow-up (No. of patients)	5	6
Mean length of follow-up (months)	18 (6-48)	19 (6-26)
Stenosis	3	1
Constipation	4	3
Incontinence	2	
PSARP Redo	2	1

as required to consent uncontaminated perineal surgery; 8 stomas were severely prolapsed and needed re-doing in four cases. In under resourced contests colostomy is performed in most cases by inexperienced health officers under local anesthesia and on the transverse colon; this exposes to a 26%-53% complication rate.[6,8,9] An added problem is the resistance of parents to stoma and lack of proper nursing care. A great deal of work was employed in parental education and supplying devices for an appropriate stoma-care. However, in our opinion the greatest effort must be put in reducing complications rate and the time of hospitalization. Prevention of perineal wound infection may be a real challenge despite accurate bowel preparation and diversion of urine stream by 5-6 days of indwelling bladder catheter. Scarce nursing staff, poor hygienic conditions, shortage of cleaning products, and abundant mucous discharge from the new anal opening all contribute to a high rate of wound infection and dehiscence. Our modified technique resulted in a significant lowering of these post-operative complications and reduced hospital stay and the need of catheterization. Stump resection, two weeks after operation, could be performed under mild sedation on day care basis and the procedure was well tolerated by our patients without any major discomfort. Cosmetic results were fairly satisfactory and there was no delay starting the dilatation protocol. Among late complications, constipation was the most difficult to manage. Whenever related to surgical failure and anal stenosis it sometimes required re operation, mainly in Group A. Unfortunately in both groups of patient, about one third of patients were lost during follow-up and another third could not be followed long enough to prevent possible anal strictures related to family's resistance to follow the dilatation protocol.

In conclusion, management of ARM in under resourced contests remains far from satisfactory. Late diagnosis, inadequate facilities, shortage of trained paediatric surgeons, and high risk of complications even in that limited number of cases who receive proper care, still make life for these children miserable. A possible contribution may be offered, as in our experience, by simple technical solutions that make specialistic paediatric surgery more sustainable and effective and reduce the need for sometimes unaffordable nursing care.

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