ORIGINAL ARTICLE

Outcome of primary posterior sagittal anorectoplasty of high anorectal malformation in well selected neonates

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Abstract

Background: Conventional posterior sagittal anorectoplasty (PSARP) for high anorectal malformation (ARM) involves initial colostomy creation with its attendant complications, but primary PSARP in neonates requires no initial colostomy.
Objectives: To report on locally adapted inclusion criteria and outcomes of primary PSARP in neonates in Benin City.
Materials and Methods: Babies who presented during the first week of life in clinically stable conditions, without cardiac anomaly, and had hemogram and blood chemistry within normal ranges, were included in this prospective study undertaken at the University of Benin Teaching Hospital in 2008-2011.
Results: Fifty children with ARM comprising 19 (38%) low/intermediate and 31 (62%) high anomalies were treated during the period. Five (10%) singletons delivered via spontaneous vaginal delivery at term. Aged at operation between two and seven (mean 4) days and comprised three males and two females (ratio 1.5:1), met the inclusion criteria for primary PSARP. The procedure was well tolerated by all the babies; oral intake was commenced on the second post-operative day with nine days median hospitalization duration. No mortality was recorded on six months to four years follow-up. Apart from minor superficial perianal surgical site infection in one baby which responded to antibiotics, no post-operative sepsis or breakdown of repair was recorded. Continence and other anal functions were found excellent using the modified Wingspread scoring during follow-up.
Conclusion: These outcomes showed that with meticulous selection, primary PSARP in neonates was feasible and safe in a developing country. Multicenter studies and long-term follow-up are advocated World-wide.

Key words: Anorectoplasty, anorectal malformation, neonates, primary, posterior, sagittal

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Introduction

Anorectal malformations (ARM) comprise of a wide spectrum of anomalies which affects both boys and girls with slightly higher prevalence in boys.[1,2] It is a common surgical problem with reported incidences ranging between one in 3500 and one in 5000 live births.[1-3] Various methods of classification and surgical correction have been described and evolved over the years.[1] A supralevator rectum with or without fistulous communication between the rectum and the urethra/bladder in males or the vagina/uterus in females is generally accepted as a high type.[1-4] Until recently, outcome of management of especially the high type remained poor globally owing to frequently associated multiple congenital anomalies and high risk of overwhelming sepsis which results in rapid deterioration in clinical parameters.[1,2] Surgical management which requires prolonged exposure to general anesthesia was compounded by presentation of the majority of affected children during the neonatal period when homeostasis is poorly developed and the baby is still recovering from the stress of delivery and is adjusting to postnatal life.[5,6] Posterior sagittal anorectoplasty (PSARP) described by Alberto Pena[1,2] is a common surgical treatment which has given excellent results in many centers.[3-9] The conventional PSARP,

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however, requires a prior temporary colostomy creation for fecal diversion.[2,3]

Colostomy creation in neonates is a less invasive procedure which requires a shorter duration of exposure to general anesthesia. The diversion of feces from anorectum and perineum protects the definitive PSARP pull-through against fecal contamination and surgical site infection.[10] This made multi-stage repair which involves initial colostomy creation followed by definitive pull-through and eventual colostomy closure the conventional approach to surgical repair of high ARM.[2,3] Apart from challenges of post-operative care, colostomy is associated with many complications such as skin excoriations, wound infection, sepsis, prolapse, fluid and electrolytes loses which are poorly tolerated by young children. These result in consequent poor acceptance by parents/caregivers especially in developing countries.[10-13] Primary PSARP is a definitive pull-through carried out in neonates without a prior colostomy creation.[7,14,15] The virtually sterile meconium during the first week of life reduces the risk of infection from fecal contamination and many centers in developed countries have recorded successes with primary PSARP in neonates.[1-3,7,14] In developing countries, however, primary PSARP for high ARM is regarded as unfeasible due to the unique challenges.[6] Hence, apart from reports on primary repair of low and intermediate ARM, not many studies have been done on primary PSARP for high ARM in neonates in this subregion.[6,16]

Over four years, locally adapted inclusion criteria for primary PSARP in neonates with high ARM were adopted in a Nigerian referral pediatric surgical center to evaluate the feasibility and safety in this subregion. The aim of this paper is to report the outcome recorded and the experiences gained, which may be useful to surgeons in similar settings.

Materials and Methods

Study design

This four year prospective study on primary PSARP in neonates with high ARM was undertaken between January 2008 and December 2011 at the University of Benin Teaching Hospital, Benin City, Nigeria. Children with ARM were referred to the pediatric surgery unit of the hospital from hospitals in Edo and neighboring states during the period. The study was commenced after ethical approval was granted by the Local Ethics Committee of the hospital. Overall, only five neonates who met the inclusion criteria had primary PSARP during the study. Neonates with high anomalies who did not meet the inclusion criteria were excluded from the study and offered the conventional treatment. Using a pre-structured form, the age, sex, mode of presentation, family history of similar lesions, findings on examinations and investigations, and duration of hospitalization were documented. During follow-up after the primary PSARP, the presence of post-operative surgical site infection, post-operative sepsis, wound breakdown, anal stenosis, fecal incontinence, constipation/diarrhea, fecal impaction, rectal prolapse or retraction, and duration of follow-up were also documented.

The diagnosis of high ARM was made using a combination of clinical and radiological assessments. Using clinical evaluations, cross table lateral decubitus X-rays (with pubococcygeal line or PC line as a guide), lower vertebral X-ray, pelvic ultrasound scan, echocardiography and CT scan, all types of anorectal anomalies were successfully diagnosed, classified, and the babies screened for other associated congenital anomalies. This made it possible for anorectal anomaly to be classified as low, intermediate or high types using the Wingspread's classification.[1,2] In the absence of electrical myostimulator, preoperative and careful tactile perianal stimulation was used to demonstrate adequacy of perineal innervations, the sling of superficial muscle complex, and mark the position for the neoanus.

Inclusion criteria

These were locally adapted before commencement of the study and included:

1. Presentation during the first week of life in clinically stable condition
2. Absence of life threatening associated congenital anomaly (especially cardiac)
3. Absence of gross abdominal distension, splinted diaphragm and/or evidence of aspiration before presentation
4. Hemogram and blood chemistry within normal ranges.

Methods

Venous assess was secured with a large bore canula, blood was grouped and cross-matched, and the procedure was performed under general anesthesia with endotracheal intubation. Urethral catheter was routinely passed to empty the bladder and also serves as a guide during dissection so as to protect against urethral injury in male babies. PSARP described by Pena was carried out while the babies were secured in a jack-knife position [Figure 1]. A 2-ml syringe vent which was removed on the sixth post-operative day was inserted and secured to the neo-anus to serve as stent and for fecal diversion [Figure 1]. Antibiotics and analgesics comprising of ceftriaxone (75 mg/kg/24 h), metronidazole (7.5 mg/kg/dose), and tramadol (0.5 mg/kg/dose) were commenced at induction of anesthesia and continued for 72 h. Oral feeds and wound inspection/warm saline irrigation were commenced on the second post-operative day. Serial anal dilatation which was demonstrated to the parents who continued with it at home was started on the seventh post-operative day. Anal dilatation was commenced earlier than standard protocol which should start at two weeks post-operative to reduce cost of hospitalization and risk of nosocomial infection in these neonates. The babies were thereafter discharged to

follow-up at the surgical outpatient clinic. The follow-up protocol was a weekly visit for one month, bi-weekly visit for two months, monthly visit for six months and bi-monthly visit thereafter. Thorough perineal/ano-rectal examination and assessment of anorectal functions measured by the modified Wingspread scoring,[7] including "excellent," "good," "fair," and "poor," were done routinely on each visit.

Statistical analysis

The data obtained were entered into Microsoft Office Excel 2007 spread sheet and analyzed as count, frequency and percentage, and presented in simple tables and figures.

Results

In total, 50 children with ARM who comprised 19 (38%) with low/intermediate and 31 (62%) with high anomalies were treated during the period. Of the 31 PSARP for high ARM, five (16%) were carried out in neonates without a prior colostomy creation and were included in the study. Late referral, severe clinical compromise, established sepsis, gross abdominal distension, splinted diaphragm, and aspiration before arrival excluded 21 (68%), while associated multiple and cardiac anomalies excluded five (16%) babies with high ARM from undergoing primary PSARP. The five neonates included were singletons delivered via spontaneous vaginal delivery at full term. They were aged at operation between two and seven days (mean 4 days) and comprised three males and two females (ratio 1.5:1). The two females and one male had rectovaginal and rectourethral (rectoprotatic bed) fistula, respectively.

No anesthesia related complication was recorded as the primary PSARP was well tolerated by all the included babies. Oral intake was commenced and tolerated on the second post-operative day, and the babies were hospitalized for between eight and 10 (median 9) days after surgery. As shown in Table 1, the children have been followed up in outpatient clinic for between six months and four years and no mortality was recorded. Apart from minor superficial perianal surgical site infection and/or inflammation which responded to antibiotics in two babies, no serious post-operative wound infection, sepsis or breakdown of the repair was recorded. Minor anal stenosis which was recorded in two babies at age nine months and one year, respectively, responded to serial anal dilatation, and they were symptom free and required no more dilatation before the second post-operative year [Figure 2]. Acceptable toilet training and bowel control for age were recorded by all the babies and no incontinence or other complications shown in the table were recorded. No patient needed re-operation and/or re-admission. The overall cost of primary PSARP for the included babies ranged between N25,000 -N30,000 compared to a range of N85,00-N95,000 for those who had conventional PSARP in the centre during the period.

Discussion

Despite the unique challenges, this study showed that with meticulous selection of patients, gentle tissue handling, careful tissue dissection and committed anesthetists, primary PSARP in neonates was feasible and safe in this setting. Although very few babies were recruited, encouraging results which could be comparable to reports[9,14,15,17] in

Table 1: Age at PSARP, hospitalization duration, post-operative follow-up, and outcomes recorded in the five neonates

Variables	Baby 1	Baby 2	Baby 3	Baby 4	Baby 5
Gestational age	38 weeks	36 weeks	40 weeks	37 weeks	36 weeks
Age at operation	2 days	2 days	3 days	6 days	7 days
Sex	M	M	M	F	F
Fistula	-	-	Rectourethral (rectoprostatic)	Rectovaginal	Rectovaginal
Perineal sensation	Excellent	Excellent	Excellent	Excellent	Excellent
Urethral/vaginal injury	-	-	-	-	-
Post-operative oral intake on	2	2	2	2	2
Post-operative admission	8	8	9	10	10
Surgical site infection	-	-	-	Superficial perianal	Perianal erythema
Sepsis	-	-	-	-	-
Breakdown of repair	-	-	-	-	-
Anal stenosis	-	-	Minor	-	Minor
Fecal continence	Excellent	Excellent	Excellent	Excellent	Excellent
Constipation/diarrhea	-	Mild diarrhea	-	-	-
Fecal impaction	-	-	-	-	-
Rectal prolapse	-	-	-	-	-
Rectal retraction	-	-	-	-	-
Post-operative fistula	-	-	-	-	-
Follow-up in years	2	0.5	4	2.5	3

PSARP=Posterior sagittal anorectoplasty

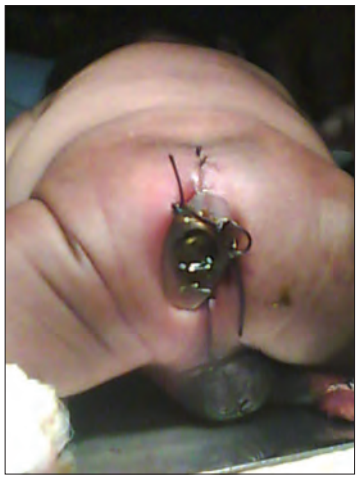

Figure 1: Immediate post-operative (primary PSARP) photograph of a 3-day-old male neonate (Baby 3) in jack-knife position with a 2-ml syringe vent in-situ for stent and fecal diversion. Note meconium efflux through the vent

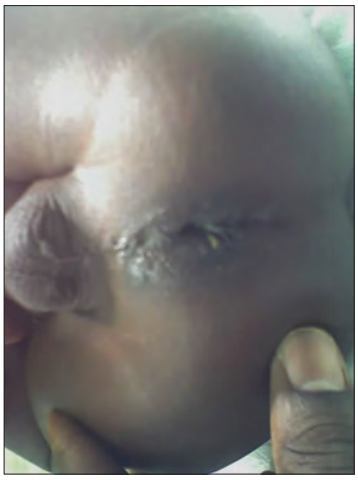

Figure 2: Left lateral positioning for perineal and anorectal examination of the same baby in Figure 1 during outpatient follow-up three years after primary PSARP

other centers were recorded in this series. Outcome of long-term follow-up reported in other studies[8,9,14,18] have not been recorded due to the shorter duration of follow-up. However, during the six months to four years follow-up, no mortality, serious surgical site infection, post-operative sepsis, breakdown of repair, rectal retraction/prolapse, post-operative recurrent fistula formation, and problematic anal stenosis were recorded, which was similar to the experiences of other authors.[7,8,14]

Additionally, follow-up assessment of post-operative functional outcome revealed that acceptable toilet training and bowel control for age were achieved after PSARP.[1-3] No incontinence, constipation, diarrhea, fecal impaction, and intractable rectal prolapse were recorded among the babies. This is unlike in other studies[9,19] where conventional PSARP in older children were associated with complications which necessitated revision surgeries. The encouraging functional outcome recorded with primary PSARP in neonates in this study could be attributed to the excellent ano-cortical connection achievable when pull-through for anorectal malformations is undertaken during the first three months of life as emphasized by earlier authors.[1-3]

The fifty children managed with anorectal malformations accounted for a large proportion of pediatric surgical workload during the period as also reported in previous similar studies in the subregion.[6,16] Of the children diagnosed with high ARM who had PSARP, only five (16%) met the locally adapted inclusion criteria for primary PSARP. The few cases included in the study were chiefly influenced by late presentation of affected babies who were in severe clinical derangements that could pose serious challenges to neonatal anesthesia in a setting with inadequate pediatric surgical facilities. The jack-knife positioning (prone and knell-elbow position with buttocks elevated) of babies during PSARP could pose an additional challenge in babies with gross abdominal distension and splinted diaphragm. Vomiting and aspiration, even with nasogastric tube in-situ, were earlier noted as major drawbacks to jack-knife positioning for PSARP.[1,2,6,16] Consequently, babies who presented after the first week of life, diagnosed with associated life threatening congenital anomaly (especially cardiac), has gross abdominal distension with splinted diaphragm, aspirated before presentation with clinical evidence of sepsis, poor renal status, and derangement in hemogram and blood chemistry, were excluded from the study and offered conventional PSARP.[2,3,13]

Centers[9,14,15,17,20] in developed countries included and reported encouraging results with much larger series. This could be because their cases presented early with majority of the babies in clinically stable conditions on arrival at their units where there were availability of sophisticated facilities and skilled manpower required for management, unlike what is obtainable in the setting described. Electrical muscle stimulator and ultrasound scan have been described and used to determine perineal innervation, adequacy of pelvic floor musculature, superficial muscle complex, and/or anal sphincter complex by other authors.[9,17,18,20,21] These were reported to help in preoperative marking of the appropriate position of the new anus and the degree of anticipated post-operative fecal continence. In the absence of electrical muscle stimulator and sophisticated ultrasound scan, careful tactile stimulation which gave acceptable results was used as a rough guide in this study. Similarly in those centers,[9,17,18,20,21] specially prepared flexible large bore anal tubes were readily available and used as vent and fecal diversion. In this study, however, improvised 2-ml syringe that was inserted in the new anus and retained by suturing it to perianal skin was removed on the sixth post-operative day. It was found adequate in the absence of preformed anal tube.

The prevalence of ARM managed in this setting during the study period was comparable to reports from other centers.[7,9,14] The duration of hospitalization was shorter and the overall cost of treatment was lower than the conventional methods.[5,9] This was a welcome relief in a setting where the majority of parents belonged to the low socioeconomic class that has difficulty in settling hospital multiple bills and cope with multiple surgeries and admissions. In addition, the avoidance of colostomy associated complications and the challenges of stoma care following the adoption of primary PSARP[10,12,13] were notable and additional advantages. However, although encouraging outcomes were recorded with primary PSARP in neonates, the minuscule number of eligible and included babies in the study, the short duration of follow-up, and the single center experience, are major limitations which may not qualify the findings for detailed statistical comparison with results from other studies.[9,14,15,17,18] The results recorded, nonetheless, may serve as impetus for multicenter studies with long-term follow-up in developing countries because findings in this study support the fact that primary PSARP is feasible and safe in awareness-poor and resources-limited regions.

Conclusion

By adopting locally adapted inclusion criteria, five out of thirty-one neonates who were treated for high ARM successfully underwent primary PSARP in the setting. Late presentation of clinically compromised babies was a major exclusion criterion which emphasizes the need for early referral of children with ARM. The outcomes recorded with primary PSARP in neonates showed that the procedure is feasible and safe in developing countries, which may serve as encouragement to surgeons in similar settings who may wish to undertake the procedure in neonates. Multicenter studies and long-term follow-up are advocated world-wide for a generally accepted conclusion on the applicability and safety of the procedure.

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