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Research Article

Anorectal Malformations in Female Patients and Results of Anal Transposition in Low/Intermediate Anomalies

Javaid Iqbal* and Nabi Bux Napar

Department of Pediatric Surgery, The Children's Hospital and the Institute of Child Health, Lahore, Pakistan

***Corresponding Author**: Javaid Iqbal, Department of Pediatric Surgery, The Children's Hospital and the Institute of Child Health, Lahore, Pakistan.

Received: October 21, 2021; **Published:** October 29, 2021

Abstract

Objectives: To analyze our experience in the treatment of low/intermediate anorectal anomalies with staged anal transposition and assess prevalence of various malformations and associated defects in female patients. Study Design: Dissertation based retrospective study.

Place and Duration of Study: This study was conducted at the Department of Pediatric Surgery The Children's hospital and the institute of child health Lahore Pakistan and Mayo Hospital/KEMC Lahore, Pakistan from January 2012 to August 2015.

Patients and Methods: During a period of two years and eight months from January 2016 to August 2015, 304 patients with various anorectal malformations were admitted to our department. These included 218 males and 86 female patients. 40 female patients presented in neonatal age. Initial procedure performed for anovestibular/rectovestibular fistula was cutback anoplasty or colostomy followed by anal transposition/PSARP at a later time. Patients with anovestibular and rectovestibular fistulae were divided at random into two groups and first group (A) underwent three staged procedure and second group(B) underwent two staged procedure. In this way management was completed in 30 patients.

Results: Male to female ratio observed in present study was 2.5:1. Anovestibular fistula (54%) was found to be most common lesion in these patients followed by rectovaginal and rectovestibular fistula (9%). Imperforate anus without fistula was present in 10 patients. Associated anomalies were found to coexist in 26(33%) patients.

Conclusion: Large majority of patients comprised of fistulous lesions in the patient population studied. Two staged Anal transposition is comparable (more advantageous) and cost effective than three staged procedure for management of these fistulous lesions. This kind of surgery should always be performed with a covering colostomy.

Keywords: *Anovestibular/Rectovestibular Fistula; Female Imperforate Anus; Anal Transposition; Treatment*

Introduction

The word malformation carries a stigma with it. That of distortion and imperfect self. Malformation of anal region always produced feeling of desperation and hopelessness. The malformation of anorectum are common congenital anomalies. The treatment of the condition has come a long way since the time of sopranos of Ephesus who recommended rupturing the anal membrane with the tip of the little finger [41]. Through ages various surgeons have strived to restore these patients to normalcy both anatomically and physiologically. Anatomical correction being easy to obtain, main challenge facing the surgeon today is to achieve continence. The understanding of embryology and anatomy of hind gut has greatly improved in recent years due to new imaging modalities like CT, MRI and other techniques

and mostly they present in early life [19]. Many of these fistulas are sufficient in size to decompress bowel except few cases when fistula dilation/cutback anoplasty [31] or colostomy is required to evacuate bowel. Anal transposition was described in 1826 and has become popular because external sphincter complex can be accurately identified with electrical stimulation and anus can be redirected through this muscle complex [35,44].

Aim of the Study

The aim of this operation is to restore the perineal body and anorectal angle while maintaining function. However, the procedure should be covered by diverting colostomy to avoid infection and dehiscence of the wound.

Patients and Methods

A total of 86 female patients with various anorectal malformations were admitted through emergency and OPD to our department. A separate registration number was allotted to each patient on admission and detailed history and physical examination was carried out followed by relevant investigations. Examination under anesthesia was carried out when required. Apart from CBC and serum electrolytes specific investigations like invertogram, ultrasound, echocardiography, CT/MRI were carried out individually as needed. For the sake of description, management of female patients with ARM have been divided into neonatal and definitive management.

Forty female neonates with various anorectal malformations were admitted during the study period. All neonates were nursed in NICU and their temperature was stabilized before any surgical procedure was performed. I/V line was secured and infusion of 0.18%. N/S in 10% D/W started. All neonates were given vit k1 in a dose of l mg intramuscularly and 0.5ml of tetanus toxoid intramuscularly. An NG tube was passed when needed to rule out tracheo esophageal fistula and to relieve abdominal distension. In cases of an vestibular fistula cutback anoplasty/dilatation was performed. In all cases having intermediate or high lesions, proximal divided sigmoid colostomy. was constructed as an initial procedure followed by definitive procedure at 6 - 12 months of age [19,21]. Broad spectrum antibiotics were started in all cases requiring colostomy before induction of anesthesia as follows:

- Inj. Ampicillin 50 - 100 mg /kg/day in 4 divided doses
- Inj. Gentamycin 3 - 5 mg /kg/day in 2 - 3 divided doses
- Inj. Metronidazole 30 mg/kg/day in 3 divided doses.

Following colostomy antibiotics were continued for a period of 3 - 5 days [42]. In cases of cutback anoplasty dilatation with. Hegafs dilator of appropriate size was started on 7* postoperative day.

Two staged anal transposition procedure included colostomy and anal transposition as first stage followed by colostomy closure 6 - 12 weeks later, where as three staged procedure included colostomy followed by anal transposition 6 - 12 weeks and colostomy closure after 12 weeks [16]. Patients with anovestibular/rectovestibular fistulae were divided at random for two or three staged anal transposition procedure [24].

In all cases undergoing two staged procedure bowel preparation was started as under:

- Low residue diet + oral antibiotics 3 days preoperatively.
- Fluid diet + oral antibiotics 2 days preoperatively.

The cumulative evaluation of functional results [1] in the management of anovestibular/rectovestibular fistulae after closure of colostomy was based on a clinical criterion classifying results into good in 14 cases (47%), fair in 10 cases (33%) and poor in 6 cases (20%). Kelly's method of assessment of continence was used to measure and describe functional results:

1. Staining or smearing:
    a. No staining 2
    b. Occasional staining 1
    c. Constant staining 0.
2. Occurrence of accidental defecation/soiling
    a. None 2
    b. Occasional 1
    c. Constant 0.
3. Strength of puborectalis muscle on digital examination
    a. Strong squeeze 2
    b. Week squeeze 1
    c. No squeeze 0.

Results

During the period of study a total of 1348 female patients with various disorders were admitted to our department. These include 294 female patients with various congenital malformations affecting gastrointestinal tract. Overall there were 218 male patients affected by same anomaly in comparison to 86 female patients. The prevalence of female patients with anorectal malformations to males is 1:2.5.

Out of these 40 presented in neonatal age. Ten patients had imperforate anus without fistula and presented with neonatal intestinal obstruction. These are earliest presentations (< 3 days old). Late presentation included an vestibular/recto vestibular fistulae (63%). The oldest patient in our study was 9 years of age (a case of an vestibular fistula). The most common presentation was abnormal perineal appearance or passage of meconium through abnormal opening in fistulous lesions.

The most common complaint of patients with an vestibular fistula was constipation relieved by enemas etc. In those cases presenting with fistulous lesions diagnosis of anorectal malformation was made on clinical examination of perineum alone.

Radiological investigations (invert grams, x-rays and ultrasound) was helpful in selected cases like imperforate anus without fistula and its complications (pneumoperitoneum) and for detection of associated anomalies as renal, esophageal, genitourinary and skeletal anomalies. Similarly, echocardiography was performed only in selected cases where cyanosis or cardiac murmur was present [12].

The distribution of various types of anorectal malformations as seen in our study [2,17] according to wingspread's classification is given in table 1 below.

Type of Anomaly	No. of Patients and % ages
Imperforate anus (high)	4 (4.6%)
Imperforate anus (low)	6 (7%)
Common cloaca	2 (2.3%)
Rectal atresia	2 (2.3%)
Rectovaginal fistula	12 (14%)
Rectovesibular Fistula	8 (9%)
Anovestibular fistula	47 (54%)
Anocutaneous fistula	22 (3%)
Anal stenosis	2 (2.3%)
Anteriorly placed anus	1 (1.1%)

Table 1: *Type of anorectal malformations in female patients according to Wingspread's classification.*

Twenty six patients (33%) were found to have various associated anomalies [2,12,16,22] which are summarized in table 2.

Associated congenital anomalies in our study are summarized in table 2.

Congenital anomalies	Total No. of patients
Gastrointestinal lesions	
TE Fistula	3
Exomphalos	1
Umbilical hernia	1
Cardiac lesions	
VSD	3
Fallot's tetrology	1
Urogenital lesions	
Hydronephrosis	4
Bicornuate uterus	1
Lymphangioma of labium majus	1
Skeletal lesions	
CTEV	9
Down's syndrome	2

Table 2: *Associated congenital anomalies in cases of patients of anorectal malformations.*

The management of 40 neonates with various anorectal malformations is summarized in table 3 [9].

Presentation	No. of Patients	Management
Imperforate anus (high)	4 (10%)	Colostomy
Rectal atresia	1 (2.5%)	Colostomy
Common cloaca	1 (2.5%)	Colostomv
Rectovaginal fistula	2 (5%)	Colostomy
Rectovestibular fistula	1 (2.5%)	Colostomy
Anovestibular fistula	24 (60%)	Cutback anoplsty
Anocutaneous fistula	1 (2.5%)	Anoplasty
Imperforate anus(low)	5 (13%)	Anoplasty
Anal stenosis	1 (2.5%	Dilatation

Table 3: *Neonatal presentation and their management in cases of Anorectal malformations in female patients.*

Neonatal presentations and their management

Anal transposition was performed in a total of 30 patients presenting beyond one year of age. These included 26 cases of anovestibular and 4 cases of rectovestibular fistulae. 16 cases of anovestibular fistulae were managed by 3 staged anal transposition whereas 10 cases were managed by 2 staged procedure. Four cases of rectovestibular fistula in which anal transposition was performed were complicated by fistula recurrence (2 cases) and rectal stump retraction in 1 case. These three cases were managed later by PSARP [32,36-39].

We performed proximal divided sigmoid colostomy in left iliac fossa in all cases of anovestibular/rectovestibular fistula. Colostomy was performed either as an initial procedure or as part-of staged management plan in cases of anovestibular/ rectovestibular fistula. Complications noticed in these cases of colostomy [28] are summarized in table 4.

Complication(s)	No. of Patients	% age
Skin excoriation, dermatitis etc.	21	70
Colostomy diarrhea	3	10
Wound dehiscence	Nil	-
Stoma prolapse	1	3
Para colostomy hernia	1	*5*

Table 4: *Complications in cases of colostomy in patients of anorectal malformation.*

Complications of colostomy in anovestibular/rectovestibular fistulae.

Mostly dermatitis was found to be associated with custom made cloth worn around waist as collecting bag and it responded to application of zinc oxide paste. Minor wound infection responded to wound care and dressings (antibiotics in occasional cases). Colostomy diarrhea was managed by oral metronidazole.

Electrostimulation of sphincter muscle complex combined with digital examination was routinely carried out prior to closure of colostomy. Complications noted in these cases of colostomy closure are given in table 5.

Complications		
Perineal wound infection	Nil	Nil
Fecal soilage	Nil	10
Fistula recurrence	Nil	3
Rectal stump retraction	Nil	1
Rectal mucosal prolapse	4	1
Neoanal stenosis	4	2

Table 5: *Complications in cases of colostomy in patients of Recto/ anovestibular fistulae.*
Total cases: 30.

Minor wound infection in cases of colostomy closure occurred in 5 cases (16%) and was managed by wound care, healing by secondary suturing. While one case of adhesive bowel obstruction settled conservatively.

Comparison of two staged and three staged anal transposition is given in table 6. Postoperative complications following anal transposition in both groups are listed. Rectal mucosal prolapse occurred in 5 cases treated by trimming of mucosa.4 cases of neoanal stenosis responded to dilatations while two needed y-v anoplasty. Three cases of rectovestibular fistula complicated by fistula recurrence and rectal stump retraction were later managed by PSARP.

Documentation	3 Staged Procedure	2 Staged Procedure
Need of bowel preparation	No	Yes
Postoperative restriction of oral feeding	No	Yes
Visits of patients to hospital	3	2
Cost effectiveness (economic burden)	More	Less
Total cases (anovestibular fistulae)	16	10
Rectovestibular fistulae	1	3

Table 6: *Comparison of two vs three staged anal transposition procedure.*

Functional results [5]:

- Good: 5 - 6 points
- Fair: 3 - 4 points
- Poor: 2 points or less
- Total patients: 30.

Good	8	6
Fair	6	4
Poor		3

It was found that continence for faeces was achieved in 24(80%) patients while two patients were incontinent of liquid/diarrheal stools. No patient was incontinent of urine while soilage occurred in 6 patients (20%) and constipation in 8(26%). All these problems were managed conservatively.

Length of stay: Length of stay was dependent on the procedure performed. In case of anoplasty/cutback average length of stay in hospital was 2 days, in case of colostomy it was 5 days, in anal transposition 7 days and colostomy closure 5 - 7 days. In complex/complicated cases where PSARP was to be performed, average length of stay was 10 days.

Mortality: There were three deaths all occurring in neonatal period [9,17]. 2 cases of high variety of imperforate anus associated with tracheo-esophageal fistula died postoperatively on 5th and 7th post operative day due to chest infection and septicemia. One case of imperforate anus (high variety) died due to septicemia because of colonic perforation.

Follow up: Mean follow up was 3 months in cases of anoplasty, 9 - 12 months in case of cutback procedure, one year for colostomy in cases of 3 staged anal transposition and 3 months in cases of 2 staged anal transpositions. Mean follow up for anal transposition/PSARP was 3 - 6 months [11,15,17].

Discussion

Anorectal malformations are among the most common congenital anomalies of gastrointestinal tract presenting to pediatric surgeons [11]. Its prevalence in total neonatal admissions (1175) to our department is 1:5. However this figure is not representative as many patients are being treated in other centers in Lahore.

Males patients (72%) dominated our series compared to females (28%). This conformed with observations of Hameed S., *et al.* (1992) [46]and Raza MN (1989) [47-49], but is at variance reported in the west [33,34].

The age at presentation is variable with majority 46(54%) of patients presenting beyond neonatal period [7]. Out of 86, a total of 40 female patients presented in neonatal age. GI obstruction was the compelling problem in these delayed presentations which can be attributed to lack of health education/health facilities available to the people as many of the deliveries in our country are conducted out of the hospital.

Majority of patients with fistulous lesions presented with complaint of constipation [11,13]. The distribution of various types of lesions was different in our series than reported in previous study done by Hameed S., *et al.* in l992. In our series high anomalies constituted 18.6% compared to 31%, intermediate anomalies constituted 9% compared to 26% whereas low anomalies are 72% compared to 43% (See table 7).

The frequency of associated malformations in present study is 33% which parallels that of Hameed S., *et al.* but is low compared to 45% (Swenson 1990) and 66% (Cook RCM, 1990) [2,8,18].

Author(s)	Total No. of patients	Male	Female
Cook RCM 1990 Royal Liverpool children's Hospital Alderhay 1953-85	535	351 (65)	184 (34.4%)
Templeton and Dietsheim 1986 Children's Hospital Philadelphia 1958-79	313	182 (58%)	131 (42%)
Mclorie., *et al*. Hospital for sick children Toronto 1963-80	484	303 (63%)	181 (37%)
Swenson 1990 Children memorial Hospital 1970-86	216	132 (61%)	84 (39%)
Raza MN., *et al.* 1989 Nishter Hospital Multanl 980-88	571	420 (74%)	151 (26%)
Hameed S., *et al*. 1991-92 Mayo Hospital Lahore	152	110 (72%)	42 (28%)
Present study Jan, 1996 to Aug, 1998	304	218 (72%)	86 (28%)

Table 7: *Anorectal malformations in various reported series.*

There were 55 cases of low/intermediate anomalies (47 anovestibular + 8 rectovestibular fistulae) in our series and operative treatment (two vs three staged procedure) was completed in 30 cases. 25 cases are at various stages of treatment plans and are not included in this study.

We performed anal transposition in 26 cases of anovestibular fistulae and found this procedure to be superior to cutback anoplasty described by Dennis Brown for this anomaly [2,4]. About 80% of patients after cutback procedure complained of constipation, poor cosmetic appearance of perineum and soilage. Incidence of constipation after anal transposition was found to be less (30%) with improved appearance of perineum and there are less complaints of ascending UTI [4].

There were 4 cases of rectovestibular fistulae in our study managed by anal transposition and complicated by rectal stump retraction and fistula recurrence in immediate postoperative period (within 10 days). Probably this was due to poor mobilization of rectum causing excessive tension at neoanus to skin anastomosis. All these cases were managed by PSARP with satisfactory results.

Two staged anal transposition procedure was found to be superior to three staged procedure as it saves the patient of:

- One general anesthesia and its attendant risks.
- At least one admission and stay in hospital along with expenses/cost involved.
- There is only one disadvantage of two staged procedure as there is need of bowel preparation, otherwise we found no significant difference between two and three staged procedure as regards results and complications.

We used Kelley's criteria to assess functional outcome in cases of anal transposition which classified results into good, fair and poor [10]. Good result means regular voluntary bowel movements, no accidents of soiling with either solid or diarrheal stools, controlled passage of flatus, rare constipation and no medication required to control or regulate defecation. The poor results include no voluntary bowel movements, passage of stools constantly without any sensation. The category of fair results falls between these two extremes. It was found that continence for faeces was achieved in 24 (80%) patients while two patients were incontinent of liquid stools. No patient was incontinent of urine while soilage occurred in 6 (20%) patients and constipation in 8 (26%). All these problems were managed conservatively [10].

Generally, cutback procedure is a safe and minor operation but the anal margin remains anterior and child may face long term complications including soilage of the vagina, UTI and posterior vaginal laceration if vaginal delivery is attempted in adulthood. Some authors reported that anterior location of the anus causes constipation that can be cured by posterior anal transfer. Anal transposition is therefore recommended to avoid such problems. Anal transposition is an operation of medium complexity with excellent cosmetic results [4] and a normally situated anal opening. The colostomy is needed and recommended to prevent infection, dehiscence and fistula recurrence in these cases of anorectal malformations.

To prevent the mobilized anorectum from receding inside and forward [6], Pena recommended anterior dissection up to a point where rectum and vagina separate and have full thickness walls [2]. Okada feels 4 - 5 cm of rectal mobilization is adequate and according to Zanotti, dissection is required up to the body of the uterus. we performed dissection up to cervix or posterior fornix and found it adequate to restore normal anatomy.

Recently there are reports of anal transposition without colostomy [1,2,4,25]. In our opinion this is hazardous as there are chances of infection and subsequent disruption of wound which may impair continence. Similarly anal transposition is performed in neonatal period in some centers [5].

We strongly recommend not to perform anal transposition in the new born period. This policy enables surgeons to perform operations when infants are 6 - 12 months old and when both vaginal and rectal walls are thicker and risk of opening rectum or vagina is minimized.

Conclusion

- The prevalence of anorectal malformations in female patients cannot be accurately estimated as most of the deliveries in our setup are conducted outside hospitals and all births are not recoded.
- The association of other congenital lesions with anorectal malformations noted in our study is comparable with study done by Hameed., *et al.* 1991-92 and M N Razal 980-88.
- Two staged anal transposition is superior to three staged procedure.
- Covering colostomy [1,2] is essential part management of anovestibular and rectovestibular fistulae.
- To achieve optimum results and avoid complications it is best to perform definitive surgery when patient is 6 months or older.

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Volume 10 Issue 11 November 2021