Cross Sectional Study of Results of Postoperative Recto-vestibular Fistula Patients

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ABSTRACT

Introduction: Recto -vestibular fistula is the most common form of Anorectal malformation in females and is an intermediate type of this disease, which is treated surgically by posterior sagittal ano-rectoplasty and Anal transposition. The current study aim was to compare the outcome of two surgical techniques used for the treatment of Anorectal malformation with recto-vestibular fistula.

Material and Methods: It was the cross sectional observational study of treatment results of Anorectal malformation with recto-vestibular fistula. One hundred and forty four female children of Anorectal malformation with recto-vestibular fistula are treated by two surgical techniques. Patients were divided into two groups based on the operative procedure they had received. PSARP Group (60 patients) and Anal transposition Group (84 patients). Forty two PSARP patients and Fifty six Anal transposition patients (total 98) attended our speciality clinic of anorectal malformations. The functional results, cosmetic appearance and complications after treatment were evaluated in patients who attended for follow up and case sheet record verification done for these patients.

Results: Forty two PSARP patients and Fifty six Anal transposition (total 98) patients were evaluated. Kelly's score of continence system used to analyse the outcome. Anal transposition Group has 95% continence rates (51.7% good, 42.8%fair, 5.3% poor continence) where as PSARP group patients have 88% continence rates (38%good, 50% fair, 12% poor continence).

Conclusion: The functional outcomes were good in both treatment groups. Anal transposition Group has slightly better continence than PSARP group patients, urinary incontinence is more in PSARP group. However, perineal scarring is more in Anal transposition.

Keywords: Anal Transposition, PSARP, Anorectal Malformation, Vestibular Anus.

INTRODUCTION

Anorectal malformations (ARMs) or An imperforate anus are birth defects in which the Anus And rectum is malformed. Anorectal malformations are among the more frequent congenital anomalies encountered in paediatric surgery, with an estimated incidence ranging between 1 in 3500 and 1 in 5000 live births[1]. Antenatal diagnosis of an isolated Anorectal malformations is very rare. Majority of the patients are diagnosed in the newborn period. Anorectal malformation are wide range of varied presentation from simple low malformations like perineal fistula which requires cut back anoplasty to high complex malformations which requires difficult and challenging staged or multi-staged operative procedures[2]. Anorectal malformation management undergone tremendous changes from last few decades from disease of fatal outcome in previous decades to almost all survivals at present. Surgical procedures of anorectal malformations changed from blind pull through to anatomical sphincter based posterior sagittal anorectoplasty, then to minimal access surgical procedures with advent of laparoscopy. But even today with availability of modern imaging techniques and other accessory gadgets which assists in identifying crucial sphincter muscle complex, total absolute urinary continence and fecal continence are far from reality. Outlook of anorectal malformation changed dramatically from gloomy holistic attitude to hopeful disease with near normal life. Our prime goal in treating these anorectal malformations is to have complete urinary control and fecal control with non-restrictive social life

Perineal fistula can be identified by direct visualization of small opening at perineum. When no opening is seen, separate the labia minora and look for separate opening at the vestibule just below vaginal opening. It may be recto-vestibular fistula or vestibular anus depending on the direction upwards and posterior or downwards and posterior direction respectively. Similarly Fourchette fistula is subtype of rectovestibular fistula, seen at fourchette with wet lining anteriorly and dry skin cover posteriorly. Cloaca is single perineal opening with small genital opening. Labia minora appear somewhat abnormal and entire perineum will have only one opening. If two opening are visible(urethra and vagina),no anal opening in female child, no meconium in genitalia seen, she may be of high anorectal malformation without fistula. High malformation without fistula happens particularly in case of down syndrome or Trisomy 21. Girl with normal vagina and urethra without anal opening[3], without meconium stain need to wait for 24 hours to identify simple, low variety of malformation like perineal fistula, which may requires only simple anoplasty[4] and can be saved from colostomy

Number of classifications are proposed. Wingspread classification (table- 1)and Pena's classification (table -2) given below.

Anorectal malformation with recto-vestibular fistula is the most common form Anorectal malformations in females and is an intermediate type of disease. In this form, the fistula opens below hymen of the vagina at the posterior fourchette and is directed posterior and upward. It is managed surgically by anal transposition or posterior sagittal ano-rectoplasty. The current study aim was to compare the outcome of two surgical techniques used for the treatment of Anorectal malformation with recto-vestibular fistula

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MATERIAL AND METHODS

One hundred and forty four female children of Anorectal malformation with recto-vestibular fistula were managed at government general hospital, Guntur, Guntur Medical College, Guntur, Andhra Pradesh, India. These Patients are treated by two surgical techniques[8]. Patients were divided into two groups based on surgical procedure they had undergone. PSARP Group (60 patients) and Anal transposition Group (84 patients), (anal transposition also synonymously known as trans-sphincter ano-rectoplasty (TSARP)). All 144 patients called for follow up and assessment. Forty two PSARP patients and Fifty six Anal transposition[5] (total 98) patients were attended our speciality clinic of anorectal malformations. The functional results, cosmetic appearance and complications after treatment were evaluated in patients who attended for follow up on call in speciality clinic for Anorectal malformations and record verification done for these patients.

We treated five hundred seventy two (572) patients of Anorectal malformation during 2007 to 2017 in our department as first admission. Out of five hundred seventy two (572) patients, three hundred ninety four patients are males (68.7%) and one hundred seventy eight patients (31.1%) are females. Various anatomical variants and their number from our department are listed in (table-3).

Female	Male
High Anorectal agenesis Rectovaginal fistula, No fistula Rectal atresia	High Anorectal agenesis Rectoprostatic fistula No fistula Rectal atresia
Intermediate Rectovaginal fistula, Rectovestibular fistula Anal agenesis	Intermediate Bulbar fistula Anal agenesis
Low Anovestibular fistula Anocutaneous fistula Anal stenosis Cloaca Rare malformations	Low Anocutaneous fistula Anal stenosis Rare malformations

Table-1: Wingspread classification (1986)

Males	Females
Perineal fistula	Perineal fistula
Rectourethral fistula	Vestibular fistula
Bulbar	Persistent cloaca
Prostatic	<3cm common channel
Recto vesical (bladder neck)	>3cm common channel
Imperforate anus without fistula	Imperforate anus without fistula
Rectal atresia	Rectal atresia

Table-2: Pena's classification 1995

Procedures done (table-4)

All patients (32/32) of High ARM or recto-vestibular fistula with difficulty for regular dilatation- PSARP was done. In 9/14 of recto-vestibular fistula with oesophageal atresia with or without TEF-PSARP was done, 2/2 of Pouch colon with recto-vestibular fistula-PSARP was done,13/14 of cloacal anomalies with recto-vestibular fistula- PSARP was done, 4 of 5 Primary PSARP was done 4. Total sixty (60) patients who underwent PSARP survived[6]. Seven patients died before definitive procedure. Anal transposition was procedure of preference by us in isolated Rectovestibular fistula and ectopic anus. Other categories mentioned above, our preference was PSARP. Out of 96 girls with isolated Rectovestibular fistula, waiting for weight gain, 12 deaths reported at newborn period due to other causes. Seventy nine Rectovestibular fistula patients and five ectopic anterior anus patients underwent Anal transposition. (Total =60+84=144)

Goal of procedure is to place bowl in centre of the sphincter complex and neo-anus should be of adequate calibre. Age related expected anal sizes are mentioned in table given below (Table- 5).

The average age for operation was 15.5 months with a range of 9 months to 22 months (table-6). (Mean 15.5 months, median 15.5 months, range 13 months)

Patients above 3 years of age were evaluated for voluntary bowl

Girls (newborn) 178/572 (31.1%) of total ARMs	number	type	procedure
	12/178 girls (6.7%)	No opening, low anomaly	Anoplasty
	3/178 girls (1.6%)	Anal stenosis low anomaly	Anoplasty
	32/178 girls (17.9%)	High ARM or with difficulty for regular dilatation	Diverting colostomy
	14/178 girls (7.8%)	High anomaly with oesophageal atresia +/-TEF	Thoracotomy repair and diverting colostomy
	2/178 girls (1.1%)	Pouch colon	laparotomy, excision of pouch colon, end colostomy(1) or Ileostomy(1)
	14/178 girls (7.8%)	cloacal anomalies	Diverting colostomy
	5/178 girls (2.8%)		Primary posterior sagittal anorectoplasty
	91/178 girls (51.1%)	Rectovestibular fistula	Regular dilatation plan till weight gain for PSARP or Anal transposition
	5/178 girls (2.8%)	Ectopic anterior anus	Regular dilatation plan till weight gain for PSARP or Anal transposition
			Procedure done are-15 anoplasty, 62 diverting colostomies/stoma, 5 primary pull through. 96 wait for weight gain10kg

Table-3: Anorectal anomalies in female Newborns in our hospital

1	Anal transposition as single stage without prior colostomy 79 of 144	79 (54.8%)	Anal transposition
2	Anal transposition Ectopic anterior anus in females as single stage 5 of 144	5 (3.4%)	Anal transposition
3	PSARP done for Rectovestibular fistula in females as second stage with prior colostomy	56 (38.8%)	PSARP
4	Primary PSARP at newborn period 4 of 144	4 (2.7%)	PSARP

Table-4: Procedure done as 2nd stage or single stage after weight gain(10kg)

Age of the patient	Hegar's dilator size
1-4 months	12 Size Hegar
4-8 months	13Size Hegar
8-12 months	14 Size Hegar
1-3 years	15 Size Hegar
3-12 years	16 Size Hegar
>12 years	17 Size Hegar

Table-5: The desirable anal/anorectal calibre with respect to age should be Hegar's dilator size

Age in months at operation	Number of patients (total =98)
9 months	7
10 months	5
11 months	7
12 months	9
13 months	12
14 months	14
15 months	13
16 months	11
17 months	8
18 months	7
19 months	2
20 months	1
21 months	1
22 months	1
(Mean 15.5 months, median 15.5 months, range 13 months)	

Table-6: Age in months at operation

Age Of the patient at follow up completed years	Number of patients (98)
4 years	9
5 years	15
6 years	18
7 years	14
8 years	11
9 years	7
10 years	9
11 years	7
12 years	3
13 years	4
14 years	1
(Range 4-14= 10, mean 9 years, median 9 years)	

Table-7: Age Of the patient at follow up in years at the time of study

control, constipation, soiling, mucosal prolapse, a misplaced neo anus, anal stenosis and perineal scarring. The mean age at evaluation was 9 years, with a range of 4 to 14 years. (Range 4-14= 10, median 9 years) (Table-7).

Forty two PSARP patients and Fifty six Anal transposition (total 98) patients were evaluated by general examination, specifically for undernourishment, poor weight gain and anaemia. Local examination

Parameter	Effect	Score
Continence		
	Normal, no soiling	2
	Occasional accidents, feces/flatus escape	1
	No control, frequent accidents	0
Staining		
	Always clean	2
	Occasional staining	1
	Always stained	0
Sphincter		
	Strong and effective squeeze	2
	Weak and partial squeeze	1
	No contraction	0

Table-8: The Kelly's score of continence

PSARP	42 patients	Kelly's score	No of patients
		5-6 3-4 0-2	16 (38%) 21 (50%) 05(12%)
Anal transposition	56 patients	5-6 3-4 01	29(51.7%) 24(42.8%) 03(5.3%)

Table-9: The Kelly's score of continence result

directed to detect anal stenosis, soiling, scarring, Prolapse of rectal mucosa, displacement of anus, per rectal examination for adequate size of anus, tone of the sphincters. Enquired about social limitations, schooling problems, urinary incontinence, constipation, personality abnormalities or psychological disturbances and future expectations. X-ray abdomen and ultrasound abdomen done as required.

Various scoring systems are available in literature. Ideal scoring system is it must be easy to remember and should not be complicated or ambiguous, able to catch minor variation of incontinence

The Kelly's scoring of continence is easy to practice in outpatient department and simple to memorise. Scoring is based on stool control, sphincter squeeze, soiling. Each carries maximum of two points and minimum of "0" points (points 0,1,2). 0 to 2- poor score, 2 to 4 – fair score,5 to 6 - good score.

The Pena's continence assessment criteria —These criteria do not award points/scores but only classify three grades of continence based on factors such as controlled of stool movement, feeling of urge, able to hold the stool. A-Soiling- I. Occasional, <2/week, no change of underwear,2. Frequent, 1/day, change of underwear sometimes 3. Constant, B- Constipation- I. Manageable with diet, II Manageable with laxatives, III Manageable with enemas, C-Urinary incontinence-I. Mild dribbling day and night, II. Complete

incontinence. 3.The Holschneider continence score[7] takes more factors into consideration than Templeton scoring and more extensive, award a score of (0-4) poor continence, fair continence (5-9) and good continence (10-14).4.Templeton score based on the following six factors- Toilet training for stool, Accidents, Extra underpants/liners, Social problems, Activity restriction, Rashes. Each factor with 0, 0.5 and 1 points, classified as poor continence (0- 1.5),fair continence (2-3.5), good continence (4-5).

The Rintal continence score[8] - based on various parameters such as hold back the stool, feeling of urge, frequency of the stool, soiling of underpants, accidental passage of stool, social life restrictions and constipation. Each factor given score of 0,1,2,3 except frequency which was given score of 0,1,2. Highest score possible is 20 and does not divide into poor, fair, good results.

STATISTICAL ANALYSIS

The obtained quantitative results were expressed as mean ± standard deviation, while qualitative variables were expressed as numbers and percentages.

RESULTS

In our institute we follow Kelly's score of continence since it is simple and easy to apply even in the outpatient department settings(table 8). Kelly's score allots points for three factors of soiling, continence and sphincter squeeze each with 0.1,2. Highest score possible is six. Lowest score is 0. Good score (5-6), fair score (3-4) and poor score (0-2). It is simple and the easy to apply even in the outpatient setting. The Kelly's score of continence result (table 9). In Anal transposition group fifty six patients, 29(51.78%) patients had good continence, 24(42.85%) patients had fair continence, 03(5.35%) patients had poor continence. In PSARP group Forty two patients, 16 (38.09%) patients had good continence, 21 (50.00%) patients had fair continence, 05(11.90%) patients had poor continence. Constipation noted in 11(26.19%) patients of PSARP group, 21(37.50%) patients in Anal transposition group Prolapse noted in 3(7.14%) patients of PSARP group, 5(8.92%) patients in Anal transposition group. Displaced appearance of Anus noted in 4(9.52%) patients in PSARP group, (posterior displacement) one (1.78%) patient in Anal transposition group (anterior displacement). Anal stenosis not admitting 16 Hegar dilator in 6(14.28%) patients of PSARP group, 9(16.07%) patients of Anal transposition group. Perineal scarring Noted in 2(4.76%) patients of PSARP group, 8(14.28%) patients in Anal transposition group Overall satisfactory result reported by parents in 30(71.42%) patients of PSARP group, 51(91.07%) patients in Anal transposition group.

Social restrictions and schooling problems noted in11(26.19%) patients of PSARP group, 5(8.92%) patients in Anal transposition group Urinary incontinence (Occasional) noted in 6(14.28%) patients of PSARP group,3(5.35%) patients in Anal transposition group

DISCUSSION

Anorectal malformation with recto-vestibular fistula is an intermediate type of this disease in females in which the fistula opens near the vagina at the posterior fourchette and is adherent to the posterior vaginal wall. Patients with recto-vestibular fistula are born with the potential of bowel control, and every effort should therefore be given to perform a successful reconstruction, preferably with a single procedure. The aim of these procedures was to pull down the rectum and create a new anus within the sphincter. MRI will be useful both preoperative and post-operative periods[9]. Preoperatively for excellent anatomical details of sphincters relevant to fecal and urinary continence, pelvic muscles, sacrum and relevant sacral nerves. Post operatively for evaluation of pulled bowl position in relation to the sphincter muscles in cases of failed surgical procedures with regards to fecal continence. Low voltage electrical muscle stimulation during operative procedure may be useful in locating the centre of the sphincters which is required to gain good fecal control. Two main approaches are Posterior Sagittal Anorectoplasty (PSARP)[10] and Anal transposition[11]. In PSARP10, full visualization of the sphincter complex and clearly showed the relationship of the rectum to the urogenital system and the surrounding structures; however, it involved division of the muscle complex, levator and external sphincter, before the re-joining. Another approach is anal transposition[11] in which vestibular fistula or vestibular anus and rectum are mobilized, transposed to normal anal position by Rerouting through centre of sphincter muscle complex. In this approach, the muscle complex, the perineal body, and the perineal skin remain intact. Other techniques like the neutral sagittal anorectoplasty (NSARP) and the transfistula ano-rectoplasty (TFARP) are minor variations of Anal transposition. Children with anorectal malformations operated with problems of incontinence may develop emotional and psychological abnormalities which requires special attention from parents teachers and healthcare professionals.[13] These children be assessed by a diagnostic psychiatric interview, parental assessment and child self-report depressive scale. The incontinent children and adolescents were not judged

to be less well-adjusted than those with good bowel control. Children with anorectal malformations may have high incidence of social as well as behavioural abnormalities. Incidence of these problems may not related to the continence levels. Parental support immensely helpful to contain the abnormal behaviours. Early psychological evaluation will be beneficial.

Postoperative anorectal incontinence[13] needs to be investigated and suitable corrective measures to be worked out[14]. Common imaging and functional studies[14] done are endorectal ultrasound, contrast enhanced CT, MRI scan, Dynamic defecogram under fluoroscopy, functional study by manometry, electrical activity of sphincter complex by Electromyography.

CONCLUSION

The functional outcomes were good in both treatment groups. Overall continence 88% to 95%. Anal transposition Group has slightly better continence than PSARP group patients, urinary incontinence is more in PSARP group. However perineal scarring is more in anal transposition probably more wound infection. Most patients have voluntary bowel movements with minimal soiling very occasionally, depending on the efficiency in the treatment for constipation. For those patients that suffer from fecal incontinence, a bowel management program must be implemented. Not improved with good bowel management program, and are embarrassed to receive enemas, they have option of the creation of a continent appendicostomy, an operation that consists in connecting the tip of the appendix to the deepest portion of their umbilicus and plicating the cecum around the appendix to create a one-way-valve mechanism that allows the passing of a catheter to deliver an enema While sitting on the toilet.

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