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Outcome of posterior versus anterior sagittal ano-rectoplasty in patients of anorectal malformation (ARM) with vestibular fistula

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Abstract

Background:Vestibular fistula is the commonest ARM in females. Operative techniques described for treating vestibular fistula mainly include posterior sagittal anorectoplasty (PSARP) and anterior sagittal anorectoplasty (ASARP). Very few studies have been carried out to compare these two procedures. The study was aimed to compare the outcome following PSARP and ASARP for vestibular fistula.

Methods: This quasi-experimental study was performed in the Department of Paediatric Surgery, Chittagong Medical College Hospital. Twenty patients with vestibular fistula, both with and without colostomy, were subjected to anorectoplasty either through PSARP (n=10) or ASARP (n=10). Patients were followed up until 3 months. Patients of both groups were compared for perioperative and postoperative complications, cosmetic outcomes, and bowel function.

Results: The two groups were comparable regarding age, maturity at birth, clinical presentation, sacral ratio, and associated congenital anomalies. Median operation time was 112.5 min for PSARP and 140.0 min for ASARP (p=0.280). Four cases had wound infection and subsequent wound dehiscence after ASARP operation and none after PSARP operation (p=0.08). Two cases had

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mucosal prolapse after ASARP and one after PSARP (p=1.0). All patients, irrespective of the surgical approaches, had good bowel function except one patient with ASARP who developed faecal incontinence.

Conclusions: PSARP was associated with lesser complications and better cosmetic outcomes than ASARP for the treatment of vestibular fistula but these were not statistically significant.

Key words: Anorectal malformation, Vestibular Fistula, Posterior sagittal ano-rectoplasty, Anterior sagittal anorectoplasty.

Introduction

Vestibular fistula is the predominant type of ARM seen in females.Both posterior sagittal anorectoplasty (PSARP) and anterior sagittal anorectoplasty (ASARP) are now the two most commonly practiced procedures for vestibular fistula worldwide, although there are other modifications 1-3. Traditionally, the procedure is done in three stages which includes a colostomy, PSARP or ASARP and colostomy closure. Now a days, many surgeons are performing primary ASARP or PSARP without colostomy with success especially for babies less than 6 months old who are yet to weaning to solid food. This approach avoids the need for performing 3 surgeries, avoids ugly scars and complications of stoma and stoma closure and is cost effective 4-6.

The European consensus meeting of ARM network members concerning diagnosis and early management of new-borns with ARM also advocated primary repair for vestibular fistula depending on the expertise of the surgeon and general condition of the patients⁷. The American College of Surgery tele mentoring task force has recently published guidelines for surgical techniques that can be used to standardize intraoperative teaching and expectations for trainees

for PSARP for vestibular fistula⁸. However, there is scarcity of studies comparing these methods. The very few studies that compared the outcome following PSARP and ASARP, produced contradicting results and have limitations of expertise of the surgeons, done by trainees, and centres that do not deal with bulk of patients. With this background, this prospective quasi-experimental study has been designed to compare the outcome of PSARP and ASARP for vestibular fistula.

Methods

This was a quasi experimental study performed in The department of Paediatric Surgery, Chittagong Medical College Hospital, Chattogram, Bangladesh from September 2020 to August 2021. Patients with vestibular fistula were divided into 2 groups into PSARP and ASARP group. The first patient was allocated randomly by lottery and subsequent patients were placed in either procedure alternatively. Patients with gross cardiac anomalies or other major congenital anomalies who were considered unfit for anaesthesia by the anaesthesiologist, patients with associated meningomyelocele and patients with hugely dilated gut due to delayed presentation who would need abdomino-perineal anoplasty were excluded from the study. The research hypothesis was "PSARP has less per-operative and post-operative complications than ASARP" and the objectives were to assess peroperative complications between two groups, specifically vaginal injury and compare the postoperative clinical outcomes in terms of wound related complications, anal stenosis, mucosal prolapse, sphincter tone and cosmetic outcome. PSARP was performed as described by Levitt & Peña,9-11 and ASARP was performed as described in the study by Saoji and Nagdeve¹². Cases were prepared by rectal washout for 24 hours through the fistula, and a single

dose of third-generation cephalosporin was given 12 hours before surgery. Patients were kept on intravenous fluids for five postoperative days in cases of primary anoplasty to minimize the soiling of the wound in either group, and patients with a prior stoma will have oral feeding six hours postoperatively. The follow-ups were done at one week, two weeks, four weeks, and three months. Categorical data were expressed as frequency (percentage). Quantitative data were expressed as median and interguartile range. The proportion was compared between two groups using Fisher's exact test, and median values were compared between two groups by Mann-Whitney U test. P<0.05 was considered statistically significant. Prior approval was obtained from the ethical and review committee of Chittagong Medical College for conducting this study. Consent was obtained from the parents or the legal relatives of the patients after clarifying study details.

Results

20 patients were enrolled and underwent surgery in either approach (PSARP=10 and ASARP=10). Age ranged from 2 months to 2 years and both groups were comparable in terms of their median age (median 4.3 vs 6.5 months respectively, p=0.28) and age category (age fewer than 6 months in 6 patients in both groups, p=0.68). Two patients had prior colostomy in both groups. Sacral ratio was 0.72 vs 0.76, respectively, p= 0.05 (Table I). One patient in each group was preterm and others were term babies. Two patients in both groups had associated cardiac anomaly. One had small ASD Secondum, and the other had TOF with left renal agenesis in PSARP group and one had ASD Secondum and vaginal agenesis and the other had patent foramen ovale and polydactyly of left hand in ASARP group.

Table-I Baseline clinical characteristics between two groups					
Characteristics	Study gr	Study groups			
	PSARP (n=10)	ASARP (n=10)			
Preterm	1 (10.0)	1 (10.0)	1.0*		
Absent anal mark	2 (20.0)	0 (0)	0.474^{*}		
Weight at admission, kg	5.0 (4.4-6.5)	5.5 (4.8-7.3)	0.645 [†]		
Absent anal pit	0 (0)	0 (0)	NA		
Flat buttock	1 (10.0)	0 (0)	1.0*		
Shallow buttock crease	1 (10.0)	0 (0)	1.0*		
Colostomy	2 (20.0)	2 (20.0)	1.0*		
Sacral ratio	0.72 (0.68-0.73)	0.76 (0.68-0.78)	0.052^{\dagger}		

Data were expressed as frequency (%) or Median (IQR). PSARP: Posterior sagittal anorectoplasty; ASARP: Anterior sagittal anorectoplasty; †Independent-Samples Mann-Whitney U Test; * Fisher's Exact Test.; NA: Not applicable.

Table II Comparison of per-operative findings between two groups					
Findings	Study groups		P-value*		
	PSARP (n=10)	ASARP (n=10)			
Coccyx absent	1 (10.0)	1 (10.0)	1.0		
Deficient muscle complex	2 (20.0)	0 (0)	0.474		
Poor sphincter contraction	2 (20.0)	0 (0)	0.474		
Absent vagina	0 (0)	1 (10.0)	1.0		
Vaginal wall injury	1(10.0)	2 (20.0)	1.0		
Per-operative fecal soiling	1 (10.0)	1 (10.0)	1.0		

Data were expressed as frequency (%); PSARP: Posterior sagittal anorectoplasty; ASARP: Anterior sagittal anorectoplasty; * Fisher's Exact Test

Per-operatively, faecal soiling occurred respectively in one case in each group (Table II). Vaginal wall injury was observed in one and two patients respectively in PSARP and ASARP groups. Deficient muscle complexes and poor sphincter contraction were noted only in the PSARP group in two patients. However, none of these differences reached statistical significance (p > 0.05).

The Median interquartile range (IQR) duration of operation was 112.5 (86.3-137.5) minutes and 140.0 (105.0-165.0) minutes, respectively, in the PSARP group and ASARP group (P=0.28). When analysed among all patients of both groups, age of the patients correlated positively with the duration of operation (P=0.013, Pearson test) meaning that younger patients needed less operating time. However, in between group comparison, this correlation was significant for PSARP group (P=0.023) but not significant in ASARP group (P=0.309). The median (IQR) length of hospital stay following surgery was higher in ASARP group [9.0 (8.0-10.5) days] than in the PSARP group [9.5 (8.0-16.8) days], (P=0.58). Four

cases in the ASARP group developed wound infection, and all developed wound dehiscence (Table III). None of the patients in PSARP group developed wound infection. One patient had anal retraction and required colostomy. Four patients (40%) developed anal excoriation in the ASARP group compared to one (10%) case in the PSARP group. Regarding mucosal prolapse, one patient in PSARP group and 2 in ASARP group developed mucosal prolapse. In the ASARP group, two developed anal stenosis.

The cosmetic results of the perineum were assessed at each follow-up postoperatively in all cases with an arbitrary score ranging from 3 to 0. At the 3 month follow up, excellent cosmetic figures were 90% and 60% in the PSARP and ASARP groups without any statistical difference (p=0.117). Bowel function was assessed in patients without colostomy by asking the mother about bowel habits and incontinence. Bowel moved daily in the entire studied patients and there was no constipation, however, one patient in ASARP group developed faecal incontinence and needed colostomy.

Table III Postoperative complications stratified by two groups					
Complications	Study groups		P-value*		
	PSARP (n=10)	ASARP (n=10)			
Wound dehiscence	0 (0)	4 (40.0)	0.087		
Anal retraction	0 (0)	1 (10.0)	1.0		
Perianal excoriation	1 (10.0)	4 (40.0)	0.148		
Mucosal prolapse	1(10.0)	2 (20.0)	1.0		
Needed colostomy after anoplasty	0 (0)	1 (10.0)	1.0		
Anal stenosis	0 (0)	2 (20.0)	0.474		

Data were expressed as frequency (%); PSARP: Posterior sagittal anorectoplasty; ASARP: Anterior sagittal anorectoplasty; * Fisher's Exact Test

Discussion

This study was conducted to determine the better techniques in terms of outcome between the PSARP and the ASARP for repairing an ARM with vestibular fistula in a paediatric surgery unit of a tertiary hospital in Bangladesh. Twenty patients were enrolled and subjected to anorectoplasty either through PSARP or ASARP approach and were followed until three months after surgery to assess surgical, immediate postoperative outcome, and bowel function. The present study demonstrated that there was no statistically significant difference in the outcome of the two procedures.

Median age was 4.3 months and 6.5 months, respectively in the PSARP group and ASARP group. A similar observation was reported by Harjai, Sethi and Chandra, where the majority of the cases were below one year of age, and by Elrouby et al. where the age of the patients at the time of the operation ranged from 1 day to 15 years but most of the patients were less than one-year-old^{6,13}. It has been suggested by some that delayed repair beyond the neonatal period is preferable for reasons, such as handling less delicate tissue in the infant and a larger anatomic area on the perineum, which allows easier placement of the neoanus precisely in the centre of the sphincter complex¹⁴. On the other hand, with increasing age operating time became longer in the current study, meaning that it was more difficult to dissect and separate the vagina from the anorectum as the age of the patient increased. Moreover, ages of all the three patients in whom vaginal injuries occurred during separation were at least one year or more.

Adequate separation of the vagina from the rectum is the most important aspect of the anorectoplasty for vestibular fistula. It is claimed that the anterior sagittal approach gives a better view of the anterior dissection where the separation of the vagina and rectum takes place under direct vision. On the other hand, the posterior approach provides a better view of the posterior dissection of the rectum. Adeniran faced no difficulties in separating the rectum from the vagina among four girls with an ARM with a vestibular fistula during one-stage PSARP¹⁵. A similar observation of no significant injury to the rectum or vagina during surgical procedure was also reported for the ASARP technique by Zamir & Rasool¹⁶. In the current study, in the PSARP technique, 1 (10%) patient had a minimal vaginal wall injury that was repaired by a

single stitch, and 2 (20%) had vaginal wall injury during ASARP operation that required multiple stitches. A previous study from Bangladesh also reported a higher rate of vaginal injury during operation in the ASARP group (30.4%)¹⁷. However, none of the vaginal injuries in the present study developed further complications such as rectovaginal fistula. Similarly, Negm et al. reported the rectum was mobilized and separated from the vagina in all cases with ASARP, except two cases where the vaginal injury occurred, which was immediately repaired, with no subsequent complication¹⁸. In the comparative study by Hairjai et al. vaginal injury was reported in 3 out of 15 patients in the ASARP group and 2 cases out of 12 cases in the PSARP group¹³.

The present study demonstrated a statistically non-significant shorter median time for operation in the PSARP than the ASARP group. Khan et al. ¹⁷ reported mean ± SD duration was 84.3±7.12 minutes in ASARP. In Negam et al.'s study, the mean operative time was 109.61 minutes for ASARP¹⁸. In another study, the mean operative time was 105 (±15) minutes for primary ASARP¹⁹. Shehata reported the operative time ranged between 45 and 140 min (median time 80 minutes) in the ASARP group as compared to the PSARP group, where operative time ranged between 54 and 130 min (median time 80 min)⁵.

The median hospital stay following surgery was higher in the ASARP group and due to their wound infection rate, which necessitated longer in-hospital management. The mean hospital stay was eight days (ranging from 3 to 21 days) in another study conducted ASARP in 157 patients 19 . Khan et al. reported mean hospital stay was 7.9 ± 2.3 days in the ASARP group 17 . In the study of Ngam et al. 18 , oral feeding was started on the third postoperative day after ASARP, and hospital stay ranged from 4 to 6 days, except in complicated cases, where it extends up to 10 days.

Wound infection and wound dehiscence are potential complications of anorectoplasties. Wound infection of the posterior sagittal incision is very uncommon in the immediate postoperative period⁹. All the patients in the present study were examined daily in the postoperative period to see whether wound infection was present or not; it was found that no patients had wound infection in PSARP operation. On the other hand, 4(40%) patients with ASARP operations had wound infections. These four patients had partial wound disruption, which healed with conservative

treatment except one, who needed colostomy for anal retraction. They were kept for more days in the hospital and local wound care was given. Another study reported that two cases in each group developed wound infection, out of which one developed complete wound dehiscence¹³. This higher rate of wound infection in the ASARP group agreed with the previous finding of Khan et al.¹⁷, where the rate was 47.8%. Post-operative infection rate varied widely among studies. The infection rate was 11% in the ASARP group and 26% in the PSARP group in the study of Shehata⁵. Zamir et al. reported an infection rate of 7% in his series and 16% in Aziz et al. following the ASARP technique²⁰⁻²¹. Zamir & Rasool reported that superficial wound infection was noted in 12/70 (17.14%) patients in their ASARP series¹⁶. These inconsistencies in the findings could be explained by the small sample size of the mentioned studies.

It has been suggested that wound contamination could be minimized by aggressively cleansing the bowel by pre-operative total bowel irrigation and keeping the patient nil per oral for the first five postoperative days²². Similarly, the use of perioperative therapeutic antibiotics seems to be associated with decreased wound infection. In the current study, to reduce the chance of postoperative wound infection, cases were prepared by rectal washout for 24 hours through the fistula. A single dose of third-generation cephalosporin was given 12 hours before surgery. Patients were kept on intravenous fluids for five postoperative days in cases of primary anorectoplasty to minimize soiling of the wound in either group, and patients with a prior stoma had oral feeding six hours postoperatively. The wound was cleaned with saline and povidone-iodine every 12 hours, and after each passage of stool, local ointment was applied to the perineum.

One case (10%) in PSARP group developed mucosal prolapse, compared to 2 in ASARP group. The patient in the PSARP group who developed mucosal prolapse had absent coccyx, flat buttock, shallow gluteal crease, poor muscle complex and poor per operative sphincter contraction and is now being followed up. One patient in ASARP group underwent rectal mucosectomy 2 months after surgery. The reported incidence in other studies was 3.8% after PSARP²³, 8% after ASARP in a study by Afzal et al²⁴. and five (13.15%) cases developed mucosal prolapse in another study by Negm et al¹⁸. Belizon et al. stated the incidence of rectal prolapse after PSARP is

relatively low²³. In this study, anal dilatation was started on the 14th postoperative day and continued for three months by the parents with regular follow-up supervision. Although, two patients (20%) in the ASARP group presented with anal stenosis, these did not mandate revision anoplasty and improved with regular dilatation for three months. Many authors reported the development of anal stenosis owing to absence or irregular or poor compliance on regular dilatation²⁴⁻²⁶.

In the PSARP group, excellent cosmetic results were noted in 9 out of 10 patients compared to 6 out of 10 patients in the ASARP group. This can be related to the higher wound complications in ASARP group. On the other hand, in another study, cosmetic satisfaction was higher in the ASARP group with a median score of 3 compared to 2 in the PSARP group without any statistical significance⁵.

Postoperative continence is considered to be the cornerstone of any repair of ARM. All patients in both groups, except one in ASARP group, had voluntary bowel movements and remained continent whole over the day. The 4 patients with colostomy were not evaluated for continence. The single case in ASARP group who had faecal incontinence underwent extensive dissection, developed post-operative wound infection and dehiscence. Another patient with ASARP also developed urinary incontinence due to extensive dissection. Occasional attacks of soiling are very common in the initial phase of anorectoplasty and even in the long term, a large number of patients have occasional soiling. Along-term follow up study among 60 patients who had ASARP in India, 30% had occasional soiling²⁷. A nationwide, treatmentindependent assessment of faecal incontinence in patients with anorectal anomalies in Germany found that 76% of the 21 patients who underwent surgery for vestibular fistula had some form of faecal soiling²⁸. In their series, the rate of different grades of soiling was 73% among all types of ARM. Atwenty-two-yearssingle-centre experience of 594 cases of ASARP in Egypt found that 24% of the patients had some form of soiling⁶.

Similarly, constipation was not reported in any case in the study. The decreased incidence of constipation in the present study may be owing to shorter duration of follow up. Elrouby et al. reported a statistically significant relationship between the development of postoperative constipation and the age at repair being

less evident in patients who had been repaired at a younger age. This is consistent with the theories of early use of neoanus to train perineal musculature, synapses, and neuronal networks of brain-defecation reflex²⁶. However, long term follow-up at least up to the time these patients are correctly toilet trained (about three years) is needed before assessing the functional results. The oldest patient in this study had not reached an age of three years in the last follow-up. Shehata reported that there was no significant difference regarding the functional outcome between both groups in his series.

Levitt and Peña reported that PSARP provides excellent exposure of the vestibular fistula and the muscle complex and allows a precise placement of the rectum within the striated muscle complex giving a better chance to achieve continence¹¹. The separation of vagina and rectum is also easier by an upward suture traction on the anorectum and a downward counter traction in the vaginal outlet by a small mixter forceps. Other authors claimed that ASARP has many advantages over PSARP, such as an easier mobilization of rectum from vagina under direct vision and an accurate reconstruction of both sphincter muscle and perineal body^{27,29}. However, the present study demonstrated that although statistically not significant, PSARP had less postoperative complications in the management of vestibular fistula. Thus, in the light of these findings and by comparing the results of the present study to previous studies, it can be stated that there is some degree of superiority of the results of the PSARP technique when compared to the ASARP technique, not only in terms of postoperative complication but also on the level of the cosmetic outcome.

This study has several limitations. Due to COVID-19 pandemic situation, many routine surgical procedures were postponed and for this reason the sample size was small. Operations were performed by different surgeons with different level of expertise. Follow-up period is also short and all the follow ups were performed by a single observer, which is both a strength and weakness of the study. While it ensured uniformity in the observations, it could not theoretically rule out observer bias.

Conclusion

This small-scale study demonstrated that there was no statistically significant difference in outcome

between PSARP and ASARP for the surgical treatment of vestibular fistula. However, given the lower rate of complications and better cosmetic outcome, it suggests that PSARP may be advocated for treating vestibular fistula.

Conflict of Interest: None

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