Scholars Journal of Applied Medical Sciences
Abbreviated Key Title: Sch J App Med Sci
ISSN 2347-954X (Print) | ISSN 2320-6691 (Online)
Journal homepage:https://saspublishers.com

OPEN ACCESS

Pediatric Neuro Surgery

Vestibular Fistula: A Prospective Follow up Study for the Management of Anorectovestibular Fistula in Neonates

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DOI:10.36347/sjams.2023.v11i03.017 | **Received:** 24.02.2023| **Accepted:** 15.03.2023 | **Published:** 22.03.2023

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Abstract **Original Research Article**

Introduction*:** Anorectal Malformation (ARM) is one of the common congenital anomalies in the world. Incidence worldwide is 1 in 3000 to 5000 live births. Anorectal malformation is a well-recognized condition since antiquity and represents a wide spectrum of defects. Anorecto-Vestibular fistula (ARVF) is the commonest ARM in female children**.** Recto-Vestibular anus is the most common Anorectal Malformation (ARM) in female patients, in which rectum opens immediately behind the hymen in the vestibule. ***Aim of the Study*:** The aim of this study was to evaluate and compare the outcome between Trans- Fistula Anorectoplasty (TFARP) and Anterior Sagittal Anorectoplasty (ASARP) in the management of Recto-Vestibular Fistula (RVF) among neonates in a tertiary care hospital .Methods*:** This was a prospective comparative study and was conducted in the Faculty of Pediatric Surgery of Bangladesh Shishu Hospital & Institute, Dhaka, Bangladesh during the period from February 2017 to January 2021. In our study we took sixty (60) female neonates with vestibular fistula (VF). Among all neonates TFARP was done in 30 neonates and ASARP was done in 30 neonates for definitive correction of RVF. ***Result*:** In total 60 patients from both the groups completed the study. In our study we found the mean operation time in TFARP group and ASARP group was 65.97±4.63 minutes and 69.37±4.76 minutes. After operation, 60% neonates in TFARP group and 90% neonates in ASARP group developed complications. Most occurred complication was skin excoriation (20% & 26.67%) in TFARP & ASARP groups respectively. Wound dehiscence was noted significantly more in ASARP group. In TFARP group, 63.3% neonates had good, 33.3% neonates had fair and 3.3% neonates had poor functional score while in ASARP group, 56.7% neonates had good functional score & 43.3% neonates had fair functional score. ***Conclusion*:** In conclusion, we found that TFARP may provide the best postoperative cosmetic appearance, parent's satisfaction, and functional outcome, compared to ASARP. However, in the present study both TFARP and ASARP are effective in the management of recto-vestibular fistula in neonates.

Keywords: Recto-vestibular fistula, Trans-fistula Anorectoplasty (TFARP), Anterior Sagittal Anorectoplasty (ASARP).

INTRODUCTION

Anorectal Malformation (ARM) is one of the common congenital anomalies in the world [1]. Incidence worldwide is 1 in 3000 to 5000 live births [2]. Anorectal Malformation is a well-recognized condition since antiquity and represents a wide spectrum of defects [3]. ARVF is the commonest ARM in female children [4].Pena and Devries in 1982 reported Posterior Sagittal Anorectoplasty (PSARP) as an operative procedure for high or intermediate imperforate anus [5]. Procedures without colostomy have been described by different authors like anal transposition [6] repair of ARVF without opening the fourchette [7], repair of vestibular and perineal fistula, [8] Technical variations in single-stage methods have been described in different series with satisfactory

results [9-17]. A vestibular fistula is defined as an abnormal position of the anal orifice in the vestibule directly under the vaginal opening. It is the most common Anorectal Malformation (ARMs) in female children [18]. According to the ARM classification (Wingspread classification, 1984), the vestibular fistula may be an anovestibular fistula— a short fistula with a low-lying rectum—or a rectovestibular fistula—a long fistula with the rectum located at a position higher than that noted in case of the anovestibular type but still lying low [19]. To distinguish between the two types of fistulae, a probe is passed through the orifice; if it can be directed toward the coccyx, it is considered an anovestibular fistula. In contrast, if the probe only passes cranially along the posterior wall of the vagina, it is a rectovestibular fistula [20]. However, all vestibular fistulae are low anomaly according to their relationship with the pelvic floor. In the Krickenbeck classification, there is no differentiation between "rectovestibular" and "anovestibular" fistulas [21]. Recto-vestibular anus is the most common anorectal malformation (ARM) in female patients, in which rectum opens immediately behind the hymen in the vestibule [22]. Several surgical techniques are described for its correction including cutback, anal transposition, posterior sagittal anorectoplasty (PSARP) [23]. Okada *et al.,* devised anterior sagittal anorectoplasty(ASARP), where perineal skin, perineal body was cut through the midline perineal incision [24]. But Akshay*etal.,* describe newer techiniquetransfistulaanorectoplasty(TFARP) without cutting the perineal skin and perineal body [25]. The aim of this study was to evaluate the outcome of TFARP and ASARP in the management of recto-vestibular fistula in neonates.

OBJECTIVE OF THE STUDY

The main objective of the study was to evaluate and compare the outcome between TFARP and ASARP in the management of recto-vestibular fistula among neonates in a tertiary care hospital.

METHODOLOGY &MATERIALS

This was a prospective study and was conducted in the Faculty of Pediatric Neuro Surgery of Bangladesh Shishu Hospital & Institute, Sher E Bangla Nagar, Dhaka, Bangladesh during the period from February 2017 to January 2021. In our study we took sixty (60) female infants with vestibular fistula (VF). Among all neonates TFARP was done in 30 neonates and ASARP in 30 neonates for definitive correction of RVF.

These were the following criteria to be eligible for the enrollment as our study participants: a) Children aged from seven days to less than 1 month; b) Neonates with vestibular fistula (VF); c) Parents who had given consent to participate were included in the study And a) Neonates with coagulopathy; b) Neonates with previous surgical history; c) Neonates with other gross congenital anomaly; d) Parents who had not given consent for the study were excluded from our study.

All the patients were prepared preoperatively with standard bowel preparation with Erythromycin, Metronidazole; vestibular fistula was irrigated by normal saline and nothing per oral for 48 hours. Injection vitamin K was given for all neonates. Routine investigations like Hb%, TC, DC, serum electrolytes, urea, creatinine, BT/CT, Blood grouping and cross matching, Ultrasonogram of urinary system and pelvic organs. Special investigations like Echocardiography, X-ray spine were done where any abnormalities were detected on clinical examination and investigations.

Surgical Technique of Trans-Fistula Anorectoplasty (TFARP)

TFARP operation was performed under general anesthesia with caudal block. Intravenous Ceftazidime, Metronidazole and Gentamycin were given just after induction. Patients were placed in lithotomy position after catheterization. Site of the anus is marked by electrostimulation. Several fine silk traction sutures insertion around the fistula orifice. Circumferential incision was made in the vestibule. Separation of the rectum from the posterior vaginal wall was done by sharp dissection. Meticulous dissection of anorectum (about 4 to 5 cm length) was done with care not to damage the vagina or musculature enclosing the rectum (figure1). Haemostasis was ensured. Placement of mobilized rectum was performed at the proposed site of anus through centre of muscle complex then fixation of rectum to the muscle complex. Anoplasty was done by standard technique then apposition of vestibular wound.

Figure 1: Surgical technique of TFARP

Surgical Technique of Anterior Sagittal Anorectoplasty (ASARP)

ASARP operation was done under general anesthesia and caudal block. Intravenous Ceftazidime, Metronidazole and Gentamycin were given just after

induction. Patients were placed in lithotomy position after catheterization. Site of the anus was marked by electrostimulation. Several fine silk traction sutures were inserted around the fistula orifice. Circumferential incision was made in the mucocutaneous junction at the opening of fistula with posterior extension along midline to reach the centre of external sphincter muscle (anal dimple). Separation of rectum from posterior vaginal wall was done by sharp dissection. Meticulous dissection of anorectum (around 4 to 5 cm length) was done with care being taken not to cause damage to the musculature enclosing the rectum. Retro rectal division of muscle complex was performed with sparing of puborectalis muscle and electro coagulation of bleeding vessels then backward mobilization & placement of rectum was done at the centre of muscle complex (figure-2). Apposition of anterior end of fan shaped muscle and suturing by interrupted stitches were done. Fixation of rectum to the muscle over its entire circumference by interrupted stitches and anoplasty was done. Reconstruction of perineal body & apposition of vestibular and perineal wound was performed.

Figure 2: Surgical technique of ASARP

Post-Operative Management: Post-operative management was same for both surgical techniques. Post-operative treatment was as follows: paracetamol (acetaminophen) 15mg/kg /dose for pain relief in neonate and infant, Foley's catheter was kept in situ up to 5th post-operative day, Povidone iodine solution applied to the wound and at neoanus several times. Oral feeding was started on 1st post-operative day. Parenteral antibiotic continued up to 5th postoperative day. Majority of the patients were discharged on 6th postoperative day. Anal dilatation was started on 14th postoperative day with Hegar's dilator. Parents were taught to dilate the neoanus two times a day for two weeks, once daily for one month, every alternate day for one month, twice a week for one month, once a week for three months.

Follow up: Follow up schedules were 14th postoperative day, on 4th week after surgery, then 1st month, 3rd months, 6th month and then once yearly up to 2 years. If any complications developed within this period, patients were advised to attend the hospital and subsequent treatment was given accordingly.

Functional outcome was assessed by scoring system based on a personal interview with parents or guardians. This scoring system is based on a questionnaire which consists of six items. Each patient was given a score ranging from 0 to 20; 14–20 is good, 7–13 is fair, and 0–6 is poor [23].

Children were evaluated for as follows:

1. Anal position (anal position index [API] calculated by ratio of anus-posterior fourchette to coccyx-posterior fourchette distance). API <0.34 was defined as anterior displacement;
2. Anal size;
3. Facial expression during defecation;
4. Stooling frequency;
5. Soiling;
6. Perianal excoriation.

Statistical Analysis

All data regarding demographics, clinical presentation, associated anomalies, operative technique, duration of surgical time, postoperative course including complications, bowel habits and continence were recorded systematically in preformed data collection form and quantitative data was expressed as mean and standard deviation and qualitative data was expressed as frequency distribution and percentage. Statistical analysis was performed by using SPSS 23 (Statistical Package for Social Sciences) for windows version 10. Probability value <0.05 was considered as level of significance.

RESULTS

TableI: Patient's characteristics

Characteristics	TFARP (n=30)	ASARP (n=30)	P value
Termed neonate	27(90.0%)	26(86.7%)	0.688
Age at operation (in days)	14.43±5.02	17.17±5.97	0.060
Birth weight (in kilograms)	2.78±0.28	2.71±0.24	0.251
Associated anomaly	7(23.3%)	9(30.0%)	0.599

TableII: Comparison of operation time and post-operative complication

Variables	TFARP (n=30)	ASARP (n=30)	P value
Mean Operation time (minutes)	65.97±4.63	69.37±4.76	0.007*
Post-operative complications	18(60%)	27(90%)	0.100

*Statistically significant value

TableIII: Comparison of major post-operative complications

Complications	TFARP (n=30)	ASARP (n=30)	P value
Wound dehiscence	1(3.3%)	3(10%)	0.044*
Wound infection	3(10%)	3(10%)	0.421
Skin excoriation	6(20%)	8(26.67%)	0.012*
Prolapse	0	3(10%)	0.076
Retraction	2(6.7%)	1(3.3%)	0.543
Vaginal tear	2(6.7%)	3(10%)	0.688
Anal stenosis	3(10%)	4(13.3%)	0.688
Anal displacement	1(3.3%)	2(6.7%)	0.640

*Statistically significant value

TableIV: Result of functional outcome score

Functional score	TFARP (n=30)	ASARP (n=30)	P value
Good	19(63.3%)	17(56.7%)	0.472
Fair	10(33.3%)	13(43.3%)	
Poor	1(3.3%)	0(0.0%)	

Table I shows the characteristics of our study patients. Among the 60 neonates most of them (90% & 86.7%) were termed neonates In TFARP and ASARP group respectively. Age at operation was 14.43±5.02 days and 17.17±5.97 days in TFARP and ASARP group respectively. In TFARP group, 23.3% and in ASARP group, 30.0% neonates had associated anomalies.

Table II compared the operation time & complications between two groups. We found the mean operation time in TFARP group and ASARP group was 65.97±4.63 minutes and 69.37±4.76 minutes respectively and this difference of time was statistically significant between groups. After operation, 60% neonates in TFARP group and 90% neonates in ASARP group developed complication which was not statistically significant.

Table III showed the post-operative complications of both groups. Most occurred complication was skin excoriation (20% & 26.67% in TFARP & ASARP group respectively. In TFARP group, other complications that occurred in neonates were wound infection (10.0%), wound dehiscence (3.3%), retraction (6.7%), vaginal tear (6.7%), anal stenosis (10%) and displaced anus (3.3%). In ASARP group, other complications that occurred in neonates were wound infection (10.0%), wound dehiscence (10.0%), prolapse (10.0%), retraction (3.3%), vaginal tear (10.0%), anal stenosis (13.3%) and displaced anus (6.7%). Complication was noted significantly more in ASARP group compared to TFARP group.

Table IV showed the functional outcome score of both groups. Results of the functional score showed that in TFARP group, 19(63.3%) neonates had good, 10(33.3%) neonates had fair and 1(3.3%) neonates had poor functional score. On the other hand, in ASARP group, 17(56.7%) neonates had good functional score whereas 13(43.3%) neonates had fair functional score. This difference was statistically not significant.

DISCUSSION

The major objectives in the management of ARM are the relief of intestinal obstruction if present, restoration of anorectal continuity with optimal sphincter function, early postnatal establishment of the brain-defecation reflex, reduction of the physical and psychological stress to the patient and his family [26]. Several surgical techniques were described for correction of VF. However, wide range of patients still suffers different functional sequelae as constipation, soiling, and even incontinence [23].

ASARP was introduced by Okada in 1992 for treatment of rectovestibular and anovestibular fistula [24]. The advantages of ASARP are: separation of posterior vaginal wall from rectum, which is considered the most important step of the operation, takes place under direct vision; the rectum is placed and anchored within the muscle complex and the perineal body is accurately reconstructed. There is also comfortable position of the patient and operative surgeon with good

anatomical orientation during procedure [27]. TFARP is a simple surgical procedure that does not divide the levator muscle or the perineal body. Preservation of these structures contributes significantly toward improvement of the aesthetic appearance of the perineum and of fecal continence [25]. The present study evaluated the outcome of TFARP and ASARP in the management of recto-vestibular fistula in neonates.

Among the 60 neonates most of them were termed neonates. Age at operation was 14.43±5.02 days and 17.17±5.97 days in TFARP and ASARP group respectively [Table I]. Previous studies conducted among vestibular fistula patients in the Bangladesh Shishu Hospital & Institute also found that the mean age of the patients were below one month [28, 29]. In TFARP group, 23.3% and in ASARP group, 30.0% neonates had associated anomalies [Table I]. Important associated anomalies include genitourinary defects, which occur in approximately 50% of all patients with anorectal malformations [3]. As the study participants were selected purposively, the percentages of associated anomalies were found less in the present study.

The mean operation time in TFARP group and ASARP group was 65.97±4.63 minutes and 69.37±4.76 minutes respectively and this difference of time was statistically significant between groups [Table II]. The study of Khan *et al.,* also found that the mean operation time was significantly shorter in TFARP group [29].

After operation, 60% neonates in TFARP group and 90% neonates in ASARP group developed complication which was not statistically significant [Table II]. Most occurred complication was skin excoriation in both groups. This high incidence of skin excoriation was due to the stool contact with the surrounding skin. High incidence of skin excoriation was found also in other study (23.3%) [23].

Wound infection was present in 10.0% patients in both groups. This result was consistent with other study where in TFARP group, 10.0% patients developed wound infection [29].

Partial wound dehiscence was noted significantly more in ASARP group (10.0%) than TFARP group (3.3%). The cross sectional comparative study of Khan *et al.,* found more wound dehiscence in ASARP group (21.7%) than TFARP group (5.0%) [29].

Vaginal tear occurred in 6.7% and 10.0% patients in TFARP and ASARP respectively.Those vaginal tear were repaired during operation, none of them developed further complication i.e. fistula. High incidence of vaginal tear was found in the study of Khan *et al.,* as the surgeons were beginners at the time of study [29].

In TFARP group, no patient developed prolapse and 6.7% patients developed retraction. Here the percentage of retraction was more as rectum was not fixed in the muscle complex, only anus was fixed in the proposed anal position. Other study also did not find any case of prolapse after TFARP [25]. In ASARP group, 10.0% patients developed prolapse and 3.3% patients developed retraction. This might be due to straining during defecation in spite of adequate mobilization and fixation.

Anal stenosis was found in 13.3% patients and 10.0% patients in ASARP and TFARP group respectively. The study of Elsawaf and Hashish also found 20.0% patients developed stenosis after ASARP [23]. The dissimilarity of result might be due to the age variation among patients. In the present study all the patients were from seven days to one month age group. But in the study of Elsawaf and Hashish, age range was 2-4 months. The study of Mitul*et al.,* found 5.1% patients developed stenosis after TFARP [28].

Mild anal displacement was found in 3.3% patients in TFARP group and 6.7% patients in ASARP group. These did not require a redo surgery. After ASARP, other study also found 3.3% patients had mild anal displacement [23].

For evaluating functional score children were evaluated for anal position, anal size, facial expression during defecation, stooling frequency, soiling and perianal excoriation. Results of the functional score showed that in TFARP group, 63.3% neonates had good, 33.3% neonates had fair and 3.3% neonates had poor functional score. On the other hand, in ASARP group, 56.7% neonates had good functional score whereas 43.3% neonates had fair functional score [Table IV]. There was no significant statistical difference between the two groups regarding functional score. Studies regarding TFARP did not evaluate the functional score by the above mentioned system but they showed good or excellent anorectal functional in their study [28, 30]. Similar result was found in the study of Elsawaf and Hashish who evaluated the functional score of patients underwent ASARP by the same scoring system. They found majority of their patients (50%) had good score and 46.7% had fair score [23].

Constipation was found less in our study group as age of the patients were less than one month in both groups and there was no dilatation of rectum due to fecal impaction. Those who had constipation were treated with diet modification, laxative and proper toilet training.

Limitations of the Study

Our study was a single centre study. We could only study a few complications because of our short study period & limited resources. There are more

complications like insomnia, fatigue, nausea, constipation needs to be evaluated. After evaluating once those children we did not follow-up them for a long term and have not known other possible interference that may happen in the long term with these children.

CONCLUSION AND RECOMMENDATIONS

In this study we evaluated the outcome between TFARP and ASARP among neonates with VF. Though we found complications like wound dehiscence, wound infection, skin excoriation, prolapse, retraction, vaginal tear, anal stenosis & anal displacement in both groups. TFARP may provide the best postoperative cosmetic appearance, parent's satisfaction, and functional outcome, compared to ASARP. However, in the present study both TFARP and ASARP are effective in the management of recto-vestibular fistula in neonates. So, further study with a prospective and longitudinal study design including larger sample size needs to be done to identify more complications and evaluate the efficacy of TFARP and ASARP in the management of recto-vestibular fistula.

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