# Short-term outcome of one-stage sphincter-saving anterior sagittal anorectoplasty in vestibular and perineal fistulae in female infants

Mohamed Ahmed Negm[a], Mohamed Ahmed Arafa[b], Khalid Mohamed Elshimy[b]

[a]Pediatric Surgery Unit, Qena Faculty of Medicine, South Valley University, Qena, [b]Pediatric Surgery Unit, Faculty of Medicine, Tanta University, Tanta, Egypt

Correspondence to Dr. Mohamed Ahmed Negm, MD, Pediatric Surgery Unit, Qena Faculty of Medicine, South Valley University, Qena, 83523, Egypt. Tel (office): +20965211281; fax: +20965211279; ORCID iD: https://orcid.org/0000-0003-0633-9147. Official e-mail: drmohamednegm@med.svu.edu.eg, e-mail: drmohamednegm@yahoo.com

**Received:** 8 September 2019
**Revised:** 7 October 2019
**Accepted:** 27 October 2019
**Published:** 14 February 2020

**The Egyptian Journal of Surgery** 2020, 39:199–205

**Background/aim**
Vestibular and perineal fistulae are the most common anorectal malformations in female infants. The progress in surgical correction has been aimed at achievement of both good continence and cosmetic outcome. This study aimed to present two-center experiences in one-stage sphincter-saving modification of anterior sagittal anorectoplasty for vestibular and perineal fistulae in female infants.

**Patients and methods**
This prospective study was conducted on 38 patients. They were divided into vestibular and perineal groups. The position of anus was marked by Peña stimulator, and traction suture were placed around the fistula. With longitudinal midline perineal incision, separation of the rectum from posterior vaginal wall. The center of anal sphincter muscle complex was identified and the rectum was passed through it, finally anoplasty with reconstruction of perineal body. Continence was evaluated by Krickenbeck and Holschneider scores in patients older than 3 years.

**Results**
The mean age was 60.26 days. The mean operative time was 109.61 min. The rectum was passed through the intact sphincter in all cases. A total of three (7.89%) cases developed superficial wound dehiscence that was treated conservatively, one (2.63%) case needed colostomy and anal dilatation was done for 3 months, one (2.6%) case developed anal stenosis corrected by anoplasty, and five (13.15%) cases developed minor mucosal prolapse, with spontaneous improvement. Constipation occurred in six (15.78%) patients and was controlled by diet modification and laxatives. Good continence was obtained.

**Conclusion**
One-stage sphincter-saving anterior sagittal anorectoplasty is safe and feasible technique in vestibular and perineal fistulae in female infants. The short-term outcome show good cosmetic and functional results regarding continence.

**Keywords:**
anterior sagittal, fistula, one stage, perineal, sphincter saving, vestibular

Egyptian J Surgery 39:199–205

1110-1121

## Introduction

Rectovestibular fistula is the most common type of rectal malformations in newborn girls [1] Posterior sagittal anorectoplasty (PSARP) was introduced by Pena and Devries in early 1980s [2] and anterior sagittal anorectoplasty (ASARP) was introduced Okada *et al*. [3] for treatment of rectovestibular and anovestibular fistula. Regarding the perineal fistula, according to Peña, PSARP is the definite approach, and anoplasty, cutback, and ASARP with or without colostomy are the reported techniques [4,5].

In both ASARP and PSARP procedures, the sphincter muscle complex must be incised, and this potentially decreases the continence mechanism. Many modifications of ASARP such as neutral sagittal anorectoplasty [6] and transfistula anorectoplasty [7] have been advocated. Sphincter-saving technique is another modification aiming for improving the continence mechanism [8,9]. The aim of this study is to present two-center experiences in sphincter-saving ASARP in both vestibular and perineal fistulae. The feasibility of the technique, wound healing, and continence assessment using both Krickenbeck and Holschneider continence scores [10,11] in patients older than 3 years and were compared in both groups.

## Patients and methods

This prospective study was conducted on 38 female infants with vestibular and perineal fistulae in two Pediatric Surgery Unit, Qena University Hospital and Pediatric Surgery Unit, Tanta University

Hospital, Egypt, during the period from January 2014 to April 2019. The study began after obtaining ethical committee approval. Detailed informed consent form was signed by the parents or guardians of the children after explaining the hazards and possible complications of the procedures.

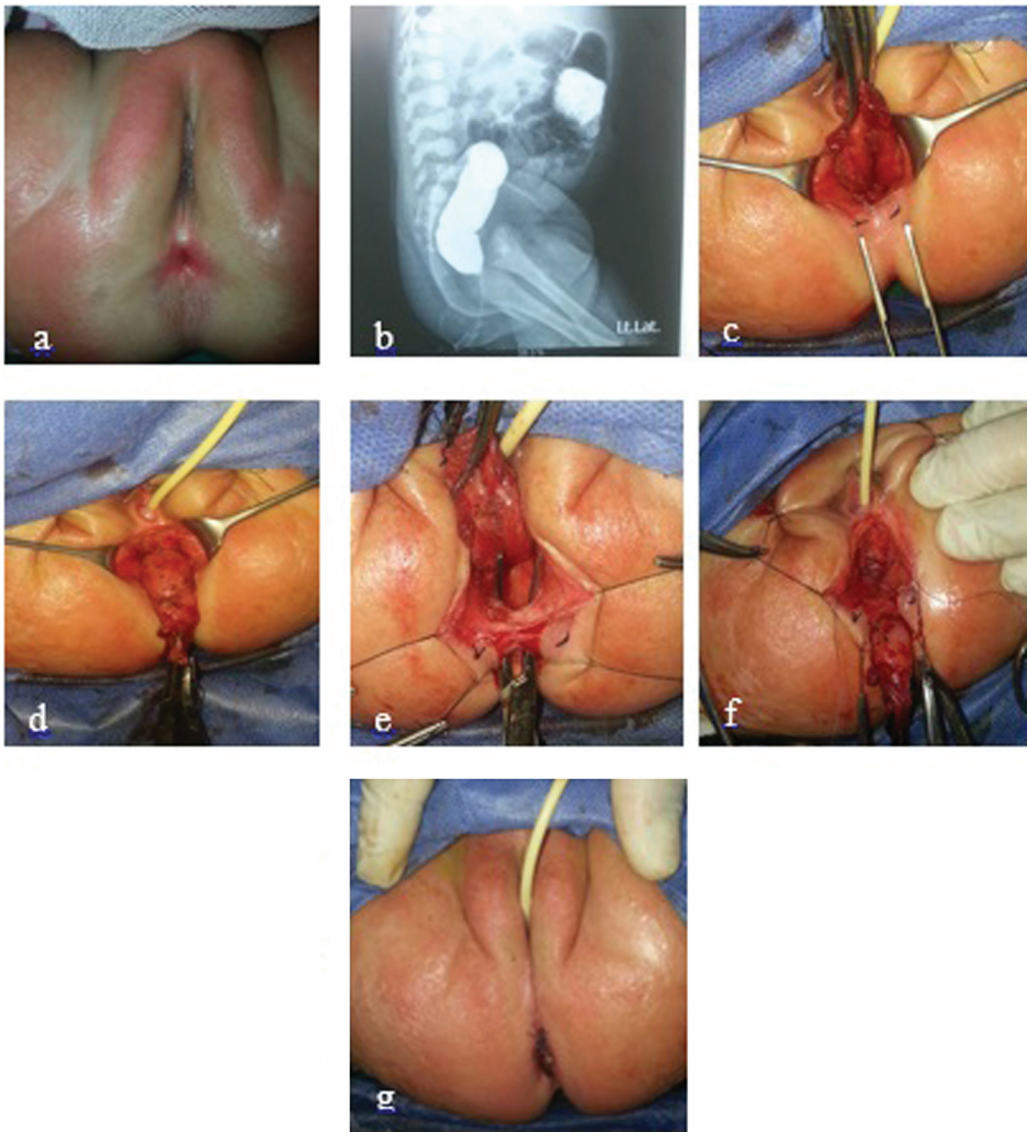
Regarding the plan of the study, the patients were grouped into two groups: group 1 included female infants with vestibular fistula, and group 2 included female infants with perineal fistula.

Proper history taking and examination, laboratory investigation, and contrast study through the fistula were done for all patients to exclude associated anomalies. It is our preference to operate after the neonatal period, and the parents were instructed for regular dilation of the fistula when needed.

Regarding the surgical technique, the surgical steps are the same in both groups except in separation of the rectum from the vagina in vestibular fistula; it needs meticulous dissection.

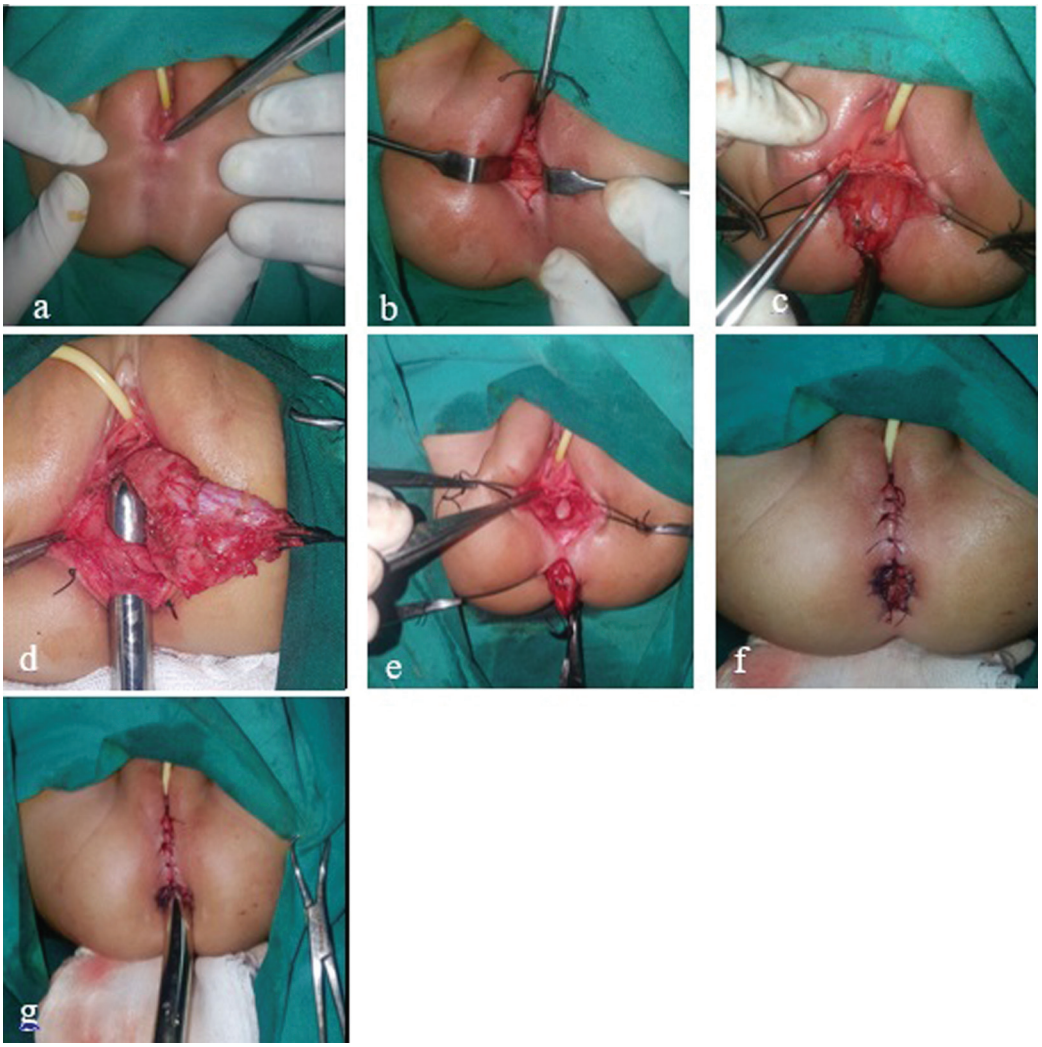
Regarding colonic preparation, the day before surgery, saline washout through the fistula was done and feeding was stopped as long as required for anesthesia. Under general anesthesia and caudal block in some cases, the patient was positioned in a supine position with legs hanging and supported. After

**Figure 1**




Operative steps in perineal fistula. (a) Preoperative. (b) Lower contrast enema through the fistula. (c) Dissection of the fistula after marking the anterior limit of the sphincter complex. (d) The rectum completely mobilized and reaching the perineum without tension. (e) Passing artery forceps through the center of muscle complex (intact sphincter). (f) Passing the rectum through muscle complex and starting reconstruction of perineal body. (g) The end of procedure by anoplasty and perineal skin closure.

**Figure 2**




Operative steps in vestibular fistula. (a) Patient with vestibular fistula. (b) Traction suture around the fistula and starting perineal dissection. (c) Mobilization of the rectum after separation from the vagina. (d) Gradual dilatation of sphincter ring using Hegar dilator (in another patient for demonstration). (e) Passing the rectum through the sphincter muscle complex. (f) The end of surgery by anoplasty, perineal body reconstruction, and skin closure. (g) Calibration of the neoanus by Hegar dilator.

urethral catheterization, the position of the neoanus was marked at the center of the sphincter muscle complex by muscle stimulator (Peña stimulator) or low current diathermy. After applying traction sutures around the fistulous opening with circumferential incision around it, strict midline perineal incision was done, with preservation of perineal skin over the muscle complex as much as we can (Figs 1 and 2).

Dissection for separation of the rectum was started posterior and then laterally, and lastly from posterior vaginal wall. Then mobilization of the rectum to reach the perineum without tension was done. This was followed by identification of the center of muscle complex and passing curved artery forceps with gradual dilation using Hegar probes up to 12–14 mm to stretch the sphincter ring, and then passing the rectum through the center of the sphincter with the aid of the traction sutures. Special care was taken not to damage the sphincter musculature. Anoplasty then was done with slight tension to guard against mucosal prolapse. Finally, reconstruction of perineal body was done with fixing of the anterior rectal wall with two stitches to prevent its retraction, and finally skin closure.

Oral feeding was started after 48 h, and systemic antibiotic (third-generation cephalosporin and metronidazole) was given with colonic preparation and was continued for 7 days according to the perineal wound condition, and urinary catheter removed after 5 days. All instructions of good local hygiene were given to the parents with aid of attending nursing staff.

Local wound care is extremely important and consists of washing of the perineum with normal saline after each passage of stool with local antibiotic ointment and diaper left open to avoid dermatitis. Anal dilatation was started 2 weeks postoperatively and was continued for 3 months in all cases. Follow-up visits were weekly in the first 3 months, monthly for the next 6 months, every 4 months for the next year, and then yearly.

**Table 1 The results in both groups**

| | Range | Mean±SD | *t* test | *P* value |
|---|---|---|---|---|
| Age (days) | | | | |
| Perineal fistula group | 43–75 | 62.06 ±11.11 | 0.940 | 0.339 |
| Vestibular fistula group | 45–75 | 58.81±9.55 | | |
| Weight (kg) | | | | |
| Perineal fistula group | 3.2–5 | 4.07±0.63 | 0.053 | 0.819 |
| Vestibular fistula group | 3.2–5.5 | 4.02±0.61 | | |
| Operative time (min) | | | | |
| Perineal fistula group | 50–80 | 64.71±7.39 | 15.854 | 0.001* |
| Vestibular fistula group | 75–100 | 87.38±5.62 | | |
| Hospital stay (days) | | | | |
| Perineal fistula group | 4–10 | 5.59±1.62 | 0.051 | 0.823 |
| Vestibular fistula group | 4–9 | 5.48±1.44 | | |
| Krickenbeck continence score[a] | | | | |
| Perineal fistula group | 6–7 | 6.64±0.50 | 0.863 | 0.363 |
| Vestibular fistula group | 5–7 | 6.38±0.77 | | |
| Holschneider continence score[b] | | | | |
| Perineal fistula group | 11–14 | 12.82±0.98 | 0.304 | 0.587 |
| Vestibular fistula group | 9–14 | 12.46±1.94 | | |

[a]Total score=7 points. [b]Total score=14 points. *Statistical significant

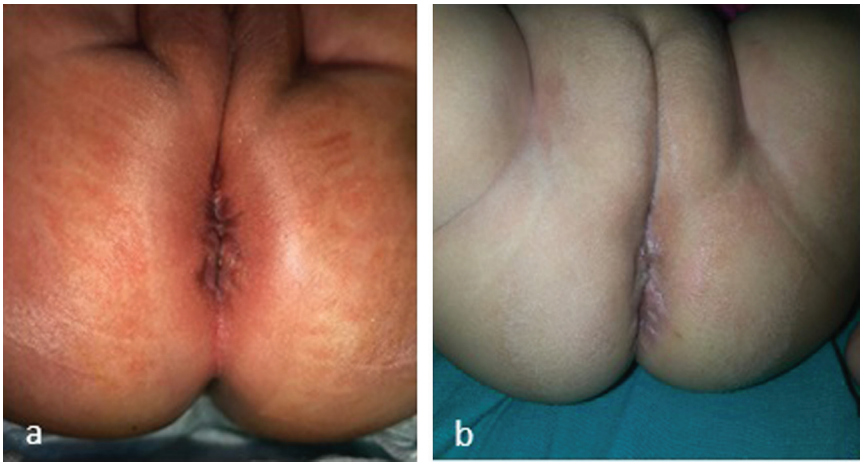
Regarding assessment of functional outcome, continence was evaluated by both Krickenbeck and Holschneider continence scores [10,11] for patients older than 3 years during follow up visits.

## Results

Among 38 infants, there were 21 (55.3%) cases with vestibular fistula in group 1 and 17 (44.7%) cases with perineal fistula. The age, weight, operative time, and hospital stay in both groups are shown in Table 1. The operative time was shorter in perineal fistula group, with statistical significance (*P*=0.001). In both groups, the rectum was mobilized and separated from the vagina, except two cases in vestibular group where vaginal injury occurred, which was immediately repaired, with no subsequent complication. In both groups, the mobilized rectum passed through the intact sphincter complex, there was no reported case of rectal injury. Oral feeding started on the third postoperative day. Hospital stay ranged from 4 to 6 days, except in complicated cases, where it extend up to 10 days.

Good wound healing was obtained in both groups (Fig. 3). Regarding wound complications (Table 2), three cases (two in vestibular fistula group and one in perineal fistula group) developed superficial wound infection that was treated with complete healing with antibiotic and daily dressing (Fig. 4). Two patients (one in each group) developed deep wound infection; the patient in perineal group needed diverting colostomy due to perianal and gluteal infection with healing by fibrosis leading to anal stenosis corrected by anoplasty before colostomy closure with good continence by the age of 3 years. A total of five (13.15%) cases developed minor mucosal prolapse, and spontaneous improvement was noticed

**Figure 3**




Follow-up at 6 months of (a) patient from perineal fistula group and (b) patient from vestibular fistula group.

after 3–6 months. Regular follow-up for anal dilatation was done for all patients for 3 months. No anterior anal migration occurred in the study cases. Constipation occurred in six patients. The follow-up period ranged from 6 to 60 months (mean±SD=36.67±17.26).

Regarding continence (Table 1), it was assessed in 24 patients (13 patients in vestibular fistula and 11 patients in perineal fistula) who became older than 3 years with maintaining regular follow-up by both Krickenbeck [10] and Holschneider [11] scores. Good results were obtained in both groups, with no statistically significant difference found. On the contrary, children who were younger than 3 years, continence was assessed by asking the mother about bowel habit and presence of soiling in between bowel motion. Sphincter tone was roughly assessed by rectal examination.

**Table 2 Operative and postoperative complications in both groups**

| Complications | *n* (%) | Group 1 (vestibular fistulae) (*N*=21) | Group 2 (perineal fistulae) (*N*=17) |
|---|---|---|---|
| Rectal injury | 0 | 0 | 0 |
| Vaginal injury | 2 (5.26) | 2 | 0 |
| Sphincter injury | 0 | 0 | 0 |
| Superficial wound infection | 3 (7.89) | 2 | 1 |
| Deep wound infection | 2 (5.26) | 1 | 1[a] |
| Diverting stoma | 1 (2.63) | 0 | 1[a] |
| Mucosal prolapse | 5 (13.15) | 4 | 1 |
| Anal stenosis | 1 (2.63) | 0 | 1[a] |
| Constipation | 6 (15.78) | 4 | 2 |

[a]The same case.

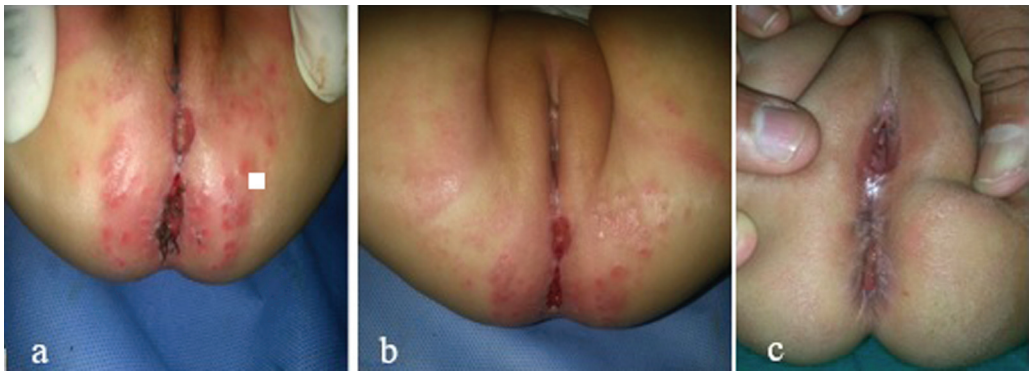
## Discussion

Imperforate anus with vestibular and perineal fistulae is the most common form of anorectal malformations in female infants [12]. The progress in the surgical techniques from cut back, V-Y plasty, PSARP, ASARP and the current discussed technique have based on understanding surgical anatomy of the perineum and the anal sphincter complex in patients with anorectal malformations. These progresses aimed to achieve both good continence and cosmetic results.

The PSARP provides excellent exposure and precise placement of the neoanal canal within the muscle complex. However, the external sphincter muscle complex and part of the lower portion of the levator mechanism need to be divided [1]. In the procedure of ASARP, only the anterior fibers of external sphincter need to be divided [6,13]. We agree with other authors that the ASARP technique have many advantages: it provides excellent perineal exposure with accurate reconstruction of the perineal body and the anterior fibers of the sphincter, it has minimal postoperative morbidity, and above all, it can be done safely during the neonatal period [3,12,14–16].

The technique of sphincter-saving ASARP maintains maximal anatomical preservation of the anal sphincter complex and physiological integrity of anal complex mechanism in continence [8,9]. In this study, the mean age was 60.26±10.27 days. Many authors prefer to correct this anomalies in neonatal periods [17,18], whereas others delay repair beyond the neonatal period for a variety of reasons such as handling less delicate tissue in the infant and a larger anatomic area on the perineum, which allows more easy placement of the neoanus precisely in the center of the sphincter complex [19]. The presented results of this study showed a good agreement with other authors

**Figure 4**




Vestibular fistula group patient (a) Superficial wound disruption and skin excoriation (due to napkin dermatitis) (b) Gradual healing and improvement (c) complete healing.

[8,17–19] that early intervention carries very good outcomes and can be done in one stage rather than staged procedure.

Oral feeding is postponed to prevent wound soiling, and in this series, oral feeding was started after 48 h. The same was reported by Upadhyaya *et al.* [17] in their series in neonates who started oral feeding after 24 h and full oral feeding after 48–72 h, but Waheeb [15] in his series started oral feeding as early as 2 h and Elsawaf and Hashish [9] in their series started oral feeding at 6–9 h postoperatively.

In the ideal practice, a protective colostomy is performed to prevent wound infection with subsequent sphincter complex dysfunction, but the colostomy itself has its own complications including peristomal excoriation, prolapse, parastomal hernia, intra-abdominal adhesions, and bowel obstruction [20,21]. In this study, all patients operated on one-stage basis, and this is the protocol followed in our centers for correction of vestibular and perineal fistula presented in infancy. The decision for colostomy was taken only in the presence of major wound complications, and this was present only in one (2.63%) patient in perineal group.

Wound healing is very important in one-stage repair. Wound complications were reported in many studies [4,12,22]. In this study, three (7.89%) cases (two in vestibular fistula and one in perineal fistula group) developed superficial wound infection and were treated conservatively with complete healing. Deep wound infection occurred in two (5.26%) cases; one case in perineal group necessitated diverting colostomy owing to perianal and gluteal infection, leading to fibrosis and anal stenosis, corrected by anoplasty before closure of colostomy. According to our experience, this technique of sphincter-saving acts as a barrier against complete wound dehiscence and anal migration. In a study by Kumar *et al.* [5], two patients in vestibular group developed total wound dehiscence with anterior anal migration, and this did not occur in our series. Moreover, in our experience, wound disruption without exposure of the rectum is not indication for colostomy, because preservation of muscle complex around the neoanus guards against complete disruption.

In this study, anal dilatation started on 15th postoperative day and continued for 3 months by the parents with regular follow-up supervision. Only one (2.63%) patient presented with anal stenosis after infection that mandate anoplasty; this low percentage may be owing to intraoperative dilatation of sphincter before passing the rectum and regular dilatation follow-up for 3 months. Many authors reported development of anal stenosis owing to absence or irregular or poor compliance on regular dilatation [5,7,13,22,23].

Regarding mucosal prolapse, five (13.15%) cases (three in vestibular fistula and one in perineal fistula group) developed minor mucosal prolapse, and spontaneous improvement was noticed after 3–6 months. The reported incidence in other studies was 3.8% after PASARP [24] and in a study by Afzal *et al.* [23], after ASARP, three cases 8% developed mucosal prolapse, with one case needing mucosectomy.

Constipation occurred in six (15.78%) patients (four in vestibular fistula and two in perineal fistula group); all of them were managed by diet modification. No fecal impaction was reported among our cases. Kumar *et al.* [5] reported constipation in 25.68% of patients, managed by rectal wash, laxatives, and diet modification, but two patients had severe recurrent constipation needed frequent manual disimpaction. Kuijper and Aronson [4] reported constipation in 34% of patients who needed regular laxative and/or enema. The decreased incidence of constipation in our study may be owing to absence of associated sacral anomalies.In this study, continence was evaluated by both Krickenbeck [10] and Holschneider [11] scores for 24 (63.15%) patients (13 in vestibular and 11 in perineal group) who become older than 3 years. Good results were obtained in all cases of both group, with no statistical significance. There were no poor results in this study, and this may be owing to preservation of sphincter anatomy together with advantage of anterior sagittal approach. Good results were also obtained after the technique of ASARP [15] using Templeton and Ditesheim [25] continence score. Other authors used different scores to assess anorectal malformation after PSARP [26]. The limitations of this study are low number of cases and lacking of long-term assessment. Moreover, the two studied groups were not equal in number owing to difference in presentation of each anomaly.

## Conclusion

Sphincter-saving ASARP is a safe and feasible technique in both vestibular and perineal fistulae in female infant. It can be safely done in one stage in early infancy. The short-term outcome shows good cosmetic and functional results regarding continence. However, more long-term studies and physiological and radiological assessments are needed.

**Financial support and sponsorship**
Nil.

**Conflicts of interest**
There are no conflicts of interest.

## References


1 Levitt MA, Peña A. Anorectal malformations. Orphanet J Rare Dis 2007; 2:1–13.

2 Peña A, Devries PA. Posterior sagittal anorectoplasty: important technical considerations and new applications. J Pediatr Surg 1982; 17:796–811.

3 Okada A, Kamata S, Imura K, Fukuzawa M, Kubota A, Yagi M, *et al.* Anterior sagittal anorectoplasty for rectovestibular and anovestibular fistula. J Pediatr Surg 1992; 27:85–88.

4 Kuijper CF, Aronson DC. Anterior or posterior sagittal anorectoplasty without colostomy for low-type anorectal malformation: How to get a better outcome?. J Pediatr Surg 2010; 45:1505–1508.

5 Kumar B, Kandpal DK, Sharma SB, Agrawal LD, Jhamariya VN. Single-stage repair of vestibular and perineal fistulae without colostomy. J Pediatr Surg 2008; 43:1848–1852.

6 Dave S, Shi EC. Perineal Skin Bridge and levator muscle preservation in neutral sagittal anorectoplasty (NSARP) for vestibular fi stula. Pediatr Surg Int 2005; 21:711–714.

7 Pratap A, Yadav RP, Shakya VC, Agrawal CS, Singh SN, Sen R. One-stage correction of recto-vestibular fi stula by transfistula anorectoplasty (TFARP). World J Surg 2007; 31:1894–1897.

8 Wang C, Li L, Liu S, Chen Z, Diao M, Li X, *et al.* The management of anorectal malformation with congenital vestibular fistula: a single-stage modified anterior sagittal anorectoplasty. Pediatr Surg Int 2015; 31:809–814.

9 Elsawaf MI, Hashish MS. Anterior sagittal anorectoplasty with external sphincter preservation for the treatment of recto-vestibular fistula: a new approach. J Indian Assoc Pediatr Surg 2018; 23:4–9.

10 Holschneider A, Hutson J, Peña A, Beket E, Chatterjee S, Coran A, *et al.* Preliminary report on the International Conference for the Development of Standards for the Treatment of Anorectal Malformations. J Pediatr Surg 2005; 40:1521–1526.

11 Ure B, Rintala R, Holschneider A. Scoring postoperative results In Holschneider A, Hutson J, eds. Anorectal malformations in children. Heidelberg: Springer 2006. 351–359

12 Wakhlu A, Pandey A, Prasad A, Kureel SN, Tandon RK, Wakhlu AK. Anterior sagittal anorectoplasty for anorectal malformations and perineal trauma in the female child. J Pediatr Surg 1992; 31:1236–1240.

13 Upadhyaya VD, Gangopadhyay AN, Pandey A, Kumar V, Sharma SP, Gopal SC, *et al.* Single-stage repair for rectovestibular fistula without opening the fourchette. J Pediatr Surg 2008; 43: 775–779.

14 Eltayeb A. The anterior sagittal anorectoplasty for the treatment of anovestibular fistula in newborn and children. Ann Pediatr Surg 2008; 1:18–21.

15 Waheeb SM. The anterior sagittal anorectoplasty technique (ASARP) for the treatment of recto-vestibular fistulae and vestibular anus in children and neonates. Ann Pediatr Surg 2005; 1:54–58.

16 Shehata SM. Prospective long-term functional and cosmetic results of ASARP versus PASRP in treatment of intermediate anorectal malformations in girls. Pediatr Surg Int 2009; 25:863–868.

17 Upadhyaya VD, Gopal SC, Gupta DK, Gangopadhyaya AN, Sharma SP, Kumar V. Single stage repair of anovestibular fistula in neonate. Pediatr Surg Int 2007; 23:737–740.

18 Zheng S, Xiao X, Huang Y. Single-stage correction of imperforate anus with a rectourethral or a rectovestibula fistula by semi-posterior sagittal anorectoplasty. Pediatr Surg Int 2008; 24:671–676.

19 Short SS, Bucher BT, Barnhart DC, Van Der Watt N, Zobell S, Allen A, Rollins MD. Single-stage repair of rectoperineal and rectovestibular fistulae can be safely delayed beyond the neonatal period. J Pediatr Surg 2018; 53:2174–2177.

20 Patwardhan N, Kiely EM, Drake DP, Spitz L, Pierro A. Colostomy for anorectal anomalies: high incidence of complications. J Pediatr Surg 2001; 36:795–798.

21 Peña A, Migotto-Krieger M, Levitt MA. Colostomy in anorectal malformations: a procedure with serious but preventable complications. J Pediatr Surg 2006; 41:748–756.

22 Wakhlu A, Kureel SN, Tandon RK, Wakhlu AK. Long-term results of anterior sagittal anorectoplasty for the treatment of vestibular fistula. J Pediatr Surg 2009; 44:1913–1919.

23 Afzal M, Talat N, Wasti AR, Ahmed S, Mirza MB, Saleem M. The anterior sagittal anorectoplasty technique (ASARP) for treatment of female anorectal malformations: our experience. Pak Pediatr 2017; J 41:153–157.

24 Belizon A, Levitt M, Shoshany G, Rodriguez G, Peña A. Rectal prolapse following posterior sagittal anorectoplasty for anorectal malformations. J Pediatr Surg 2005; 40:192–196.

25 Templeton JM, Ditesheim JA. High imperforate anus: quantitative results of long-term fecal continence. J Pediatr Surg 1985; 20:645–652.

26 Brisighelli G, Macchini F, Consonni D, Di Cesare A, Morandi A, Leva E. Continence after posterior sagittal anorectoplasty for anorectal malformations: comparison of different scores. J Pediatr Surg 2018; 53:1727–1733.