

Investigation of the feasibility and safety of single-stage anorectoplasty in neonates with anovestibular fistula

Chiyo Shirota^a, Keisuke Suzuki^b, Hiroo Uchida^a, Hiroshi Kawashima^b, Akinari Hinoki^a, Takahisa Tainaka^a, Wataru Sumida^a, Naruhiko Murase^a, Kazuo Oshima^a, Kosuke Chiba^a, Satoshi Makita^a, and Yujiro Tanaka^a

^aDepartment of Pediatric Surgery, Nagoya University Graduate School of Medicine, Nagoya, Japan

^bDepartment of Surgery, Saitama Children's Medical Center, Saitama, Japan

Corresponding author: Hiroo Uchida, MD

Department of Pediatric Surgery, Nagoya University Graduate School of Medicine,

65 Tsurumai-cho, Showa, Nagoya 466-8550, Japan

Tel.: +81-52-744-2959

Fax: +81-52-744-2980

E-mail: hiro2013@med.nagoya-u.ac.jp

Abstract

Purpose: Anovestibular fistula (AVF) is the most common type of anorectal malformation in females. Delayed anorectoplasty with fistula dilatation is commonly performed during infancy; however, we have been actively performing anorectoplasty in neonates. We report the surgical complications and postoperative defecation function associated with single-stage anorectoplasty performed in neonates.

Methods: Patients who underwent surgery for AVF between 2007 and 2017 at two institutions were retrospectively studied. The operation time, amount of bleeding, time to start oral intake, perioperative complications, and Kelly's score were compared among patients who underwent surgery as neonates and those who underwent surgery as infants.

Results: Eighteen neonates and 17 infants underwent anterior sagittal anorectoplasty. The median operation time and time to start oral intake were significantly shorter in the neonatal group (72 min; 3 days, respectively) than in the infant group (110 min, $p = 0.0002$; 5 days, $p = 0.0024$, respectively). Postoperative wound disruption was significantly more frequent in the infant group. Of the 10 patients each in the neonatal and infant groups, there was no significant difference in Kelly's score at age ≥ 4 years.

Conclusion: Single-stage anorectoplasty in neonates with AVF can be feasibly performed and does not

impair postoperative defecation function.

Levels of evidence: III

Keywords: anorectoplasty, anovestibular fistula, neonate, defecation function

Anovestibular fistula (AVF) is the most common type of anorectal malformation (ARM) in females. Delayed anorectoplasty is commonly performed in infants, with fistula dilatation during neonatal stages. AVF is a low-type of ARM. Because patients with AVF generally have a developed levator ani muscle and a sphincter complex, a single-stage repair in the neonatal period may be an alternative treatment for AVF. We have been actively performing anterior sagittal anorectoplasty (ASARP) for AVF in the neonatal period. Few studies have compared the results of anorectoplasty in the neonatal period with those in the infant stage [1].

Here, we report the surgical complications and postoperative defecation function associated with single-stage anorectoplasty in the neonatal period compared to those of delayed repair in infants.

1. Patients and Methods

Prior to commencing this study, all protocols were approved by the ethics review board at our institutes (Approval No. 2016-0499). The medical records of patients who underwent single-stage anorectoplasty for AVF between January 1, 2007 and December 31, 2017 at Nagoya University Hospital or Saitama Children's Medical Center were retrospectively reviewed. Patients who had complications due to heart disease or immune disease were excluded.

Our ASARP procedure was as follows. All patients were given general anesthesia and placed in the lithotomy position or jackknife position for surgery. The anal site was identified using a muscle stimulator and a skin incision was made in the midline; next, a plane was dissected between the parasagittal fibers. The incision, beginning from the posterior margin of the neoanus, circumscribed the fistula at its cranial end. The fistula was freed, a sufficient length of anorectum was dissected, and the vagina was separated. The mobilized rectum was pulled down, and then anoplasty was performed [1, 2]. Neonatal ASARP was performed at Nagoya University. ASARP in the infant group was performed at the Saitama Children's Medical Center. Two authors (H.U. and Y.T.) belong to the two hospitals and confirm that very similar operative methods and postoperative management were used.

The operation time, amount of bleeding, time to resume oral intake, perioperative complications, and defecation function were compared among patients who underwent surgery as neonates and those who underwent surgery as infants. Patients aged <4 years and those with neuropathy associated with spinal cord lesions were excluded from the comparison of defecation function.

Statistical analyses were performed using Fisher's exact test for categorical variables and Wilcoxon's signed-rank test for continuous variables with Jump Pro ® 11 (SAS Institute Inc., Cary, NC, USA). A p-value of <0.05 was considered to be statistically significant.

2. Results

A total of 35 patients were included in this study. Of these, 17 patients underwent surgery as neonates (neonatal group) and 18 underwent surgery as infants (infant group). The surgical procedure was ASARP in all patients.

The median age and body weight at the time of surgery were significantly lower in the neonatal group (20 days, range: 8–30 days; 3,080 g, range: 2,352–5,412 g, respectively) than in the infant group (185 days, range: 116–279 days, $p = 0.0001$; 6,505 g, range: 5,580–8,030 g, $p = 0.0001$, respectively). These are the natural outcomes as planned methodology. The median operation time and time to start oral intake were significantly shorter in the neonatal group (72 min, range: 39–123 min; 3 days, range: 2–6 days, respectively) than in the infant group (110 min, range: 63–196 min, $p = 0.0002$; 5 days, range: 3–14 days, $p = 0.0024$, respectively). The median amount of bleeding was 0 ml/kg body weight (range: 0–7) in the neonatal group and 1 ml/kg body weight (range: 0–10, $p = 0.158$) in the infant group, which was not a significant difference.

Intraoperative complications were not observed in the neonatal group, whereas rectal injury occurred in one patient in the infant group. For postoperative complications, mucocutaneous dehiscence and skin sutures occurred in two patients in the neonatal group; one patient recovered spontaneously and the other underwent re-suturing with two stitches under local anesthesia. These two patients started dilation on postoperative days 13 and 10, started oral intake on postoperative days 2 and 3, and were discharged on postoperative days 34 and 19, respectively, suggesting that the complications did not affect any other management. Wound dehiscence occurred in seven patients in the infant group; three patients recovered spontaneously, two had to stop oral intake for approximately one week, and two required re-anorectoplasty without colostomy because of major wound disruption. Additionally, one patient in the infant group had late rectal perforation requiring anorectal tubing. Postoperative complications were significantly more frequent in the infant group (Table 1).

Table 2 shows the defecation function of patients at 4 years of age. There was no significant difference in the sacral ratio between the two groups [3]. We categorized enema usage into three groups: everyday, often

(more than once per week), and occasionally (less than once per week). Of the 10 patients in the neonatal group, 6 did not require any defecation assistance, 2 required enemas or defecation assistance 1–2 times per month, 1 needed an enema 3 times per week, and 1 needed an enema every day. Additionally, of the 10 patients in the infant group, 2 did not require any defecation assistance, 3 required enemas 1–2 times per month, 1 needed an enema 1–2 times per week, and 4 needed enemas every day. Among the 10 patients in the neonatal group and the 10 patients in the infant group currently aged over 4 years, there was no significant difference in Kelly's score (Table 2) [3].

3. Discussion

Urgent surgery for anorectoplasty is usually avoided during the neonatal period because neonates generally have weak tissues and general anesthesia is highly risky. Single-stage radical surgery for AVF is often performed in infancy if defecation can be obtained with fistula dilatation during the neonatal period [4-6]. On the other hand, low-type ARM is generally corrected by a simple anoplasty in neonates, with excellent results, as they have a well-developed levator muscle and a sphincter complex. Posterior sagittal anorectoplasty (PSARP) has become the preferred operative technique for the treatment of intermediate- or high-type ARM in infants. This technique includes the confirmation of the levator muscle and sphincter complex. After confirmation of these muscle complexes, they are divided such that the rectum is located in front of the levator muscle and within the sphincter complex. In cases of intermediate- or high-type ARM, patients often have an atrophic muscle complex. PSARP is generally performed in infants, but not in neonates, as infants are likely to have more mature tissue and an easily identifiable muscle complex. In our cases, the levator muscle and sphincter complex in AVF, being low-type ARM, could be easily identified in the neonates, and ASARP was performed without difficulty. In the neonatal period, the thick vaginal wall, which is significantly affected by maternal hormones, could be easily identified and separated from the rectal muscle during surgery. Neonates have fresh skin and tissue in the perineal area with short-term bougie-ing of the fistula. Infants with AVF always have scar tissue surrounding the fistula due to long-term bougie, and dissecting the scar tissue may result in wound infection and eventual disruption.

Upadhyaya et al. reported that single-stage repair of AVF in neonates produced good results and avoided the need for bowel preparation [1]. Wakhil et al. reported that the age when ASARP for vestibular fistula can be performed has decreased, and that babies who present early enough can be operated on in the neonatal period [2]. In our study, the operation time was significantly shorter and the amount of bleeding

was less in the neonatal group. These results suggest that ASARP is not a difficult procedure, even during the neonatal period. As the procedures were performed by multiple surgeons in this study, operative outcomes may have been different depending on the operator. Furthermore, as the chief surgeons who performed the surgical procedure and postoperative management were the same in our study, there should be minimal differences between the two hospitals. Nevertheless, differences between sites may have affected the results of perioperative complications and defecation functions, but we believe our findings should be reliable to a certain extent. The early and late complication rates in our study were low in comparison to previous reports, and defecation function results were respectable [1, 2, 3, 7]. Taken together, these findings suggest that ASARP for AVF can be performed without major complications in the neonatal period.

Kulshreshtha et al. described three different regimens for patients undergoing ASARP and reported that the highest complication rate occurred in the group that was allowed early oral intake postoperatively [4]. Other reports have also reported the need for bowel rest and preparation [7-9]. In our study, we managed oral intake only as required for anesthesia preoperatively and avoided bowel preparation. All neonatal patients received prophylactic cephalosporin for 48 h postoperatively, and the time of oral intake postoperatively was significantly shorter; however, complications were not frequent. Since oral intake varies depending on postoperative management, this comparison may not be useful as an evaluation. Similarly, Upadhyaya et al. avoided bowel preparation and allowed early feeding after surgery for patients undergoing ASARP for AVF [1]. Their report suggests that the risk of surgical infection may be lower due to lower toxic fecal flora in neonates. In past reports, perioperative complication rates for anorectoplasty for AVF were reported as 14–47% [1, 4, 7, 9, 10]. However, these reports included the neonatal period, and complication rates in the neonatal and infant periods were not reported individually. Although we could not compare the complication rate in our study with that in previously reported infant cases, the complication rate in our infant case is not extremely high compared with that reported in the literature. These reports also included neonatal surgical cases, and complications were not compared between the neonatal and the infant periods. The most important long-term outcome of this surgery is continence. The reported incidence of postoperative constipation in AVF tends to be high, ranging from 28% to 60% [7, 11-14]. In the present study, Kelly's score was used to assess continence results, and did not differ significantly between the neonatal and infant groups. Our study is subject to several limitations. Mainly, the number of cases was

small and the follow-up period was short, so the findings are not conclusive. Nevertheless, our findings support the idea that single-stage ASARP in neonates with AVF can be performed and results in good postoperative defecation function.

4. Conclusion

Single-stage ASARP for AVF can be performed even in the neonatal period. There were no significant differences in perioperative complications and long-term defecation function between neonates and infants.

Compliance with Ethical Standards : All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Disclosure of potential conflicts of interest: All the authors have no conflicts of interest.

Funding: No funding was provided for this study.

References

- [1] Upadhyaya VD, Gopal SC, Gupta DK, Gangopadhyaya AN, Sharma SP, Kumar V (2007) Single stage repair of anovestibular fistula in neonate. *Pediatr Surg Int* 23:737-740.
- [2] Wakhlu A, Kureel SN, Tandon RK, Wakhlu AK (2009) Long-term results of anterior sagittal anorectoplasty for the treatment of vestibular fistula. *J Pediatr Surg* 44:1913-1919.
- [3] Kelly JH (1972) The clinical and radiological assessment of anal continence in childhood. *Aust N Z J Surg* 42:62-63.
- [4] Kulshrestha S, Kulshrestha M, Singh B, Sarkar B, Chandra M, Gangopadhyay AN (2007) Anterior sagittal anorectoplasty for anovestibular fistula. *Pediatr Surg Int* 23:1191-1197.
- [5] Adeniran JO (2002) One-stage correction of imperforate anus and rectovestibular fistula in girls: Preliminary results. *J Pediatr Surg* 37:E16.
- [6] Kyrklund K, Pakarinen MP, Koivusalo A, Rintala RJ (2015) Bowel functional outcomes in females with perineal or vestibular fistula treated with anterior sagittal anorectoplasty: controlled results into adulthood. *Dis Colon Rectum* 58:97-103.
- [7] Kumar B, Kandpal DK, Sharma SB, Agrawal LD, Jhamariya VN (2008) Single-stage repair of

- vestibular and perineal fistulae without colostomy. *J Pediatr Surg* 43:1848-1852.
- [8] Wang C, Li L, Liu S, Chen Z, Diao M, Li X, Qiao G, Cheng W (2015) The management of anorectal malformation with congenital vestibular fistula: a single-stage modified anterior sagittal anorectoplasty. *Pediatr Surg Int* 31:809-814.
 - [9] Kuijper CF, Aronson DC (2010) Anterior or posterior sagittal anorectoplasty without colostomy for low-type anorectal malformation: how to get a better outcome? *J Pediatr Surg* 45:1505-1508.
 - [10] Harjai MM, Sethi N, Chandra N (2013) Anterior sagittal anorectoplasty: An alternative to posterior approach in management of congenital vestibular fistula. *African J Pediatr Surg* 10:78-82.
 - [11] Pena A, Hong A (2000) Advances in the management of anorectal malformations. *Am J Surg* 180:370-376.
 - [12] Aziz MA, Banu T, Prasad R, Khan AR (2006) Primary anterior sagittal anorectoplasty for rectovestibular fistula. *Asian J Surg* 29:22-24.
 - [13] Heinen FL (1997) The surgical treatment of low anal defects and vestibular fistulas. *Semin Pediatr Surg* 6:204-216.
 - [14] Bukarica S, Marinkovic S, Pekovic-Zrnic V, Dobanovacki D, Borisev V, Likic J (2004) Clinical evaluation of fecal continence after posterior sagittal anorectoplasty in anorectal abnormalities. *Med Pregl* 57:284-288.

Table 1. Comparison between the neonatal group and infant group.

	Neonatal group (n = 17)	Infant group (n = 18)	<i>p</i>
Patient characteristics			
Age at surgery (days)	20 (8–30)	185 (116–279)	0.0001*
Body weight at surgery (g)	3080 (2352–5412)	6505 (5580–8030)	0.0001*
Surgery			
Operation time (min)	72 (39–123)	110 (63–196)	0.0002*
Volume of bleeding (ml/Kg)	0 (0–7)	1 (0–10)	0.158
Time to start oral intake (days)	3 (2–6)	5 (3–14)	0.0024*
Postoperative complications			
Total	2 (13%)	7 (41%)	0.0324*
Minor wound disruption	2 (13%)	4 (24%)	
Major wound disruption	0	2 (12%)	
Late perforation	0	1 (5.9%)	

Table 2. Constipation at 4 years of age.

	Neonatal group (n = 10)	Infant group (n = 10)	<i>p</i>
Sacral ratio	0.99 (0.62–1.19)	0.88 (0.62–1.19)	0.1200
Kelly's score at 4 years of age	6 (5–6)	5 (3–6)	0.0610
Enema usage	Every day 1; ^a Often 1; Occasionally 2	Every day 4; ^a Often 1; Occasionally 3	

^aOften: more than once per week