The THAI Journal of SURGERY 2018;39:122-127.
Official Publication of The Royal College of Surgeons of Thailand

Original Article

Outcomes of the Surgical Treatment of Infants with Imperforate Anus and Vestibular Fistula without Primary Colostomy

Sujitra Kebthong, MD
Achariya Tongsin, MD
Varaporn Mahatharadol, MD
Department of Surgery, Queen Sirikit National Institute of Child Health, Bangkok, Thailand

Abstract

Introduction: **Female infants with imperforate anus with vestibular fistula can be surgically treated with either one - stage repair without colostomy or conventional multi-stage repair after primarily prospective colostomy. One stage approach is more preferable in the present period.**

Objective: **The aim of this study was to access the outcomes of a single-stage repair without colostomy in female infants with imperforate anus and vestibular fistula.**

Materials and Methods: **Medical records of the patients who were diagnosed imperforate anus with vestibular fistula and treated with a single-stage repair without colostomy during 2012 to 2016 at Queen Sirikit National Institute of Child Health were reviewed. Demographic data, operative procedures and outcomes of treatment were evaluated.**

Results: **During the study period, 34 female infants with imperforate anus and vestibular fistula underwent a single-stage repair without protective colostomy at our institute. They were classified as anovestibular fistula (AVF) in 15 cases (44.1%) and rectovestibular fistula (RVF) in 19 cases (55.9%). Most of the patients were term and normal birthweight infants. Cardiovascular defects were the most common associated anomalies. Infants with AVF were surgically treated by cut-back anoplasty (5 cases), anal transfer (4 cases) and anterior sagittal anorectoplasty or ASARP (6 cases). Infants with RVF underwent only anal transfer (5 cases) and ASARP (14 cases). Wound infection and wound dehiscence were the common postoperative complications which were noted in approximately 15% and 25%, respectively. Anorectal retraction and recurrent fistula occurred in one case, each. Both cases required redo-operation for correction several weeks later without protective colostomy. One-third of the patients had constipation with more often in RVF than AVF. Constipation was managed by toilet training, laxative and occasional enema. There was no mortality in all of the patients.**

Conclusion: **Female infants with imperforate anus and vestibular fistula could be safety treated by a single-stage repair without protective colostomy. From the present study, even major complications occurred and required redo-operation, the patients could be successfully corrected without diverted colostomy.**

Keywords: **Imperforate anus, Vestibular fistula, Single-stage repair, Without protective colostomy, Outcome**

Correspondence address: *Achariya Tongsin MD, Department of Surgery, Queen Sirikit National Institute of Child Health, 420/8 Rajavithi Road, Bangkok 10400, Thailand; Telephone/Fax: +66 234 8095, E-mail: surgeryunit@hotmail.com*

Introduction

Imperforate anus with vestibular fistula (IA with VF) is the most common form of anorectal malformations (ARMs) in female infants. The diagnosis is based on physical examination. In this condition, the fistula lies between the vagina and the fourchette, and the opening is surrounded by vestibular mucosa. According to International[2] and Wingspread's Classification[3], vestibular fistula (VF) can be classified into rectovestibular fistula (RVF) which is an intermediate defect (Figure 1) and anovestibular fistula (AVF) as a low defect (Figure 2). Historically, several operative techniques were described for VF repair including cut-back anoplasty, perineal anal transfer or transplantation, Y-V and X-Z plasty, limited posterior sagittal anorectoplasty (PSARP) and anterior sagittal anorectoplasty (ASARP)[4-7].

IA with VF can be repaired with or without a protective colostomy. Traditionally, most surgeons usually advocated a protective colostomy especially in infants with RVF, in order to prevent major postoperative complications such as surgical wound infection, wound dehiscence, anorectal retraction and recurrent fistula. Recently, more pediatric surgeons prefer to treat VF with a single-stage repair without protective colostomy both imperforate anus with AVF and RVF.

The aim of this study was to evaluate the outcomes of a single-stage repair without protective colostomy in the patients with IA and VF treated at our institute.

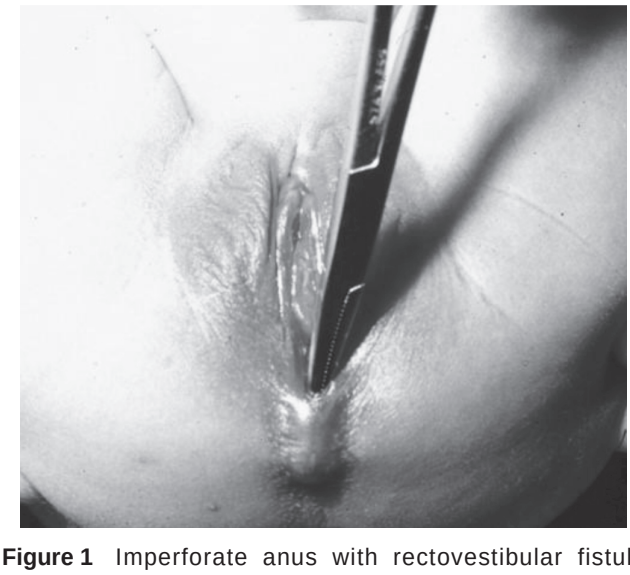

Figure 1 Imperforate anus with rectovestibular fistula (intermediate type based on International and Wingspread Classification)

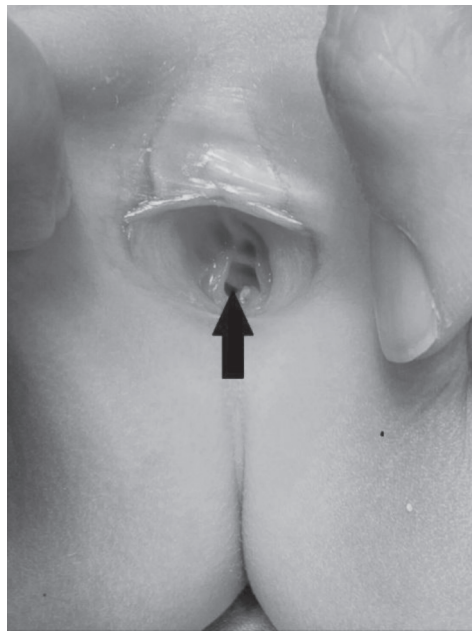

Figure 2 Imperforate anus with anovestibular fistula (low type based on International and Wingspread Classification

Materials and Methods

A retrospective study was conducted at Department of Surgery, Queen Sirikit National Institute of Child Health (QSNICH) after the proposal (Document No.61-032) had been approved by the Ethic Committees of the institute. Medical recorded of all the patients who were diagnosed IA with VF and underwent primary repair without protective colostomy at QSNCIH from January 2012 to December 2016, were reviewed. The patients who had been treated from the other hospitals or transferred for redo-surgery were excluded from the study. Demographic data, associated anomalies, type of operative procedures and results of the treatment were evaluated. Decisions for operative repair with or without protective colostomy depended on the surgeon's preference.

Data were analyzed using SPSS version 20 (IBMs® SPSS statistic). Correlations between categorical variables were evaluated by Chi-square test. A *p*-value of less than 0.05 was considered significant.

Results

During the study period, 102 female infants were diagnosed IA with VF. Only 34 cases (33.3%) were treated by a single-stage repair without protective

colostomy. According to International[2] and Wingspread's Classification[3], anovestibular fistula (AVF) : rectovestibular fistula (RVF) was noted in 15 : 19 cases (44.17% : 55.9%). Demographic data between both groups including gestational age, birth weight and associated anomalies were not different (Table 1). Most of infants with AVF underwent surgical correction in the first week of life, while those with RVF had lately repaired because RVF required fistular dilatation at home before surgery.

Infants with AVF could be treated by ASARP (6 cases), anal transfer (4 cases) and cut-back anoplasty (5 cases). Surgeons preferred correction of RVF by ASARP in 14 cases and anal transfer in 5 cases (Table 2). Cut-back anoplasty was not performed in our patients with RVF.

Eary postoperative complications were not different between AVF and RVF (Table 3). Common complications were surgical wound infection and wound dehiscence which were managed with local wound care, except wound dehiscence of 2 cases with RVF required wound re-suturing. Anorectal retraction after wound infection and dehiscence occurred in one case with RVF after ASARP and required redo-ASARP several weeks later.

Length of stay ranged from 7-31 days (average 13.3 days) in AVF and 6-45 days (average 15.3 days) in RVF. The patients were followed-up at the out-patient department 2 weeks after discharge from the hospital. Surgeons demonstrated anal dilatation with Hegar's dilator to the parents and advocated them to do with their children everyday at least 3 months. Follow-up time of all the patients ranged from 1 to 6.4 years (mean 2.64 years). Late complications included constipation and anorectal stricture occurring in both AVF and RVF. Rectal mucosal prolapse and recurrent fistula after wound dehiscence occurred only RVF, one case each (Table 3). Excision of prolapsed rectal mucosa and repair of recurrent RVF were done several weeks later. Anorectal stricture were treated by anorectal dilatation with Hegar'sdilator everyday until complete recovery. Patients with constipation were received laxative and occasional enema. The parents were encouraged to train toilet training their children at home for improvement of fecal incontinence and constipation. There was no mortality in all of our

Table 1 Demographic data of 34 female infants with vestibular fistula

Patients' data	Total No.	Anovestibular fistula Cases (%)	Rectovestibular fistula Cases (%)	*P* = value
Imperforate anus with vestibular fistula	34	15 (44.1%)	19 (55.9%)	0.304
• Gestational age (weeks) range (average)	33-40 (37.2)	36-39 (37.2)	34-40 (36.9)	0.206
• Birth weight (kg) range (average)	1.6-3.2 (2.5)	1.6-3.2 (2.5)	1.6-3.1(2.5)	0.365
• Age at operation range (average)	1 day-6.5 months (59.8 days)	1 day-5 months (37.8 days)	6 days-6.5 months (78.1 days)	0.048*
• Associated anomalies cardiovascular (PDA,ASD)	7 (20.5)	4 (26.7)	3 (15.8)	0.436
Vertebral	2 (5.8)	0	2 (10.6)	0.195
Genitourinary	1 (2.9)	0	1 (5.3)	0.863
Other	2 (5.8)	1 (6.7)	1 (5.3)	0.253

Table 2 Operative techniques for a single-stage repair without colostomy

Operative techniques	Total N = 34 cases (%)	Anovestibular fistula (N = 15) cases (%)	Rectovestibular fistula (N = 19) cases (%)	*P* = value
Cut-back anoplasty	5 (14.7)	5 (33.3)	0	0.006*
Anal transfer	9 (26.5)	4 (29.7)	5 (26.3)	0.982
Anterior sagittal anorectoplasty	20 (58.8)	6 (40.0)	14 (73.7)	0.048*

Table 3 Postoperative complications

Complications	Total N = 34 cases (%)	Anovestibular fistula (N = 15) cases (%)	Rectovestibular fistula (N = 19) cases (%)	*P* = value
Early				
• Wound infection	5 (14.7)	2 (13.3)	3 (15.8)	0.841
• Wound dehiscence	9 (26.6)	4 (26.7)	5 (26.3)	0.982
• Anorectal retraction	1 (2.9)	0	1 (5.3)	0.694
Late				
• Recurrent fistula	1 (2.9)	0	1 (5.3)	0.410
• Rectal mucosal prolapse	1 (2.9)	0	1 (5.3)	0.367
• Anorectal stricture	2 (5.8)	1 (6.7)	1 (5.3)	0.253
• Constipation	10 (29.3)	3 (20.0)	7 (36.8)	0.285

patients.

Discussion

ARMs in female neonates encompass spectrum of defects ranging from imperforate anal membrane to persistent cloaca. Imperforate anus with vestibular fistula (including AVF and RVF is the most common defect in female patients[1-3].

In the past, AVF was accepted to primary surgical correction without protective colostomy and RVF should be performed protective colostomy before definitive repair[2,3]. Nowadays, a single-stage repair without primarily colostomy for the treatment of both AVF and RVF have been advocated by many reports[5,8-12]. This approach does not only reduce the colostomy related complications, but also reduce the cost of treatment and hospital stay with comparable results[8-11].

Nevertheless, many reports provoked the importance of colostomy[6,13-16]. Without protective colostomy, wound infection and wound dehiscence are the perceived complications leading to severe stricture with fibrosis or even recurrent fistula. Furthermore, if these complications occur, a redo-surgery is known to have poorer functional outcome and the patient may have lost the best opportunity for the optimal functional results[14]. Despite the results are still debating and most authors agree that the surgeon experience is a critical factor in improving the results of a single-stage repair. Therefore, this approach should be recommended for the experienced surgeon[6,13,15].

The single-stage repair of vestibular fistula, both AVF and RVF, has been performed at our institute by some attending staffs over 10 years. Preoperative preparation, operative technique and postoperative care were individualized, depending on the surgeon's experience.

In the present study, most patients were term neonates with normal birthweight. Associated anomalies were noted in one-third of cases and cardiovascular defect was the most common anomaly, similar to the other reports[17,18].

Operative techniques for correction were chosen based on the type of fistulas and surgeon's prefer. ASARP, which was modified from PSARP by Okada[7], is the most popular method for repair of vestibular fistula, both AVF and RVF[7,13,14]. Anal transfer, which preserves the anterior part of anal sphincter, was the second frequently used in our institute. A cut-back anoplasty should not be performed in intermediate and high anomalies, such as RVF. Therefore, our patients with RVF underwent ASARP and anal transfer. A cut-back anoplasty did not used in our cases with RVF.

The most common early postoperative complications were wound infection and wound dehiscence. Wound infection occurred in our patients both AVF anal RVF with no statistic significance. The incidence of postoperative wound infection was reported ranging from 0% to 12.1%[16,18,19]. Wound dehiscence often developed after wound infection, but some cases might spontaneously occur without an evidence of infection. The incidences of wound dehiscence were reported in a wide range from 0% to

39.4%[16,18-21]. Most of wound infection and dehiscence could successfully recover by conservative treatment. Our 2 cases required re-suturing for treatment of wound dehiscence. Anorectal retraction occurred in a few cases and caused by inadequate rectal mobilization, anastomosis under tension and wound dehiscence[6]. A redo-ASARP should be done after the wound was subside from infection and inflammation several weeks. Recurrent fistula did not occur in some reports[12,16,20]. One case with RVF in this study developed a recurrent fistula with suspicion of postoperative wound infection and wound dehiscence. Repair of the recurrent fistula and redo-ASARP for management of anorectal retraction were successfully done without diverted colostomy.

Constipation was the most common late constipation in many reports[4,8,225-24]. Peña[6] reported as high as 55% of cases. The present study showed that constipation occurred in approximately 30% of all vestibular fistula and more often in RVF than AVF. Most of the patients required laxative, enema and the important toilet training from their parents.

Conclusion

By considering the disadvantages of colostomy, repair of vestibular fistula without colostomy should be a preferred and reliable approach. The outcomes were promising and none of the patients in this study required colostomy, even in the cases with having major complications.

REFERENCES

1. Peña A. Imperforate anus and cloacal malformations. In: Ashcraft K, ed. Pediatric surgery. 3rd ed. Philadelphia: Elsevier; 2000. p. 473-49.
2. Stephens FD, Smith ED. Anorectal malformations in children. Chicago: Year Book Medical Publishers; 1971.
3. Stephens FD, Smith ED, Paul NW. Anorectal malformations in children: update 1988. New York: Alan R Liss; 1988.
4. Heinen FL. The surgical treatment of low anal defects and vestibular fistulas. SeminPediatr Surg1997; 6:204-16.
5. Demirbilek S, Atayurt HF. Anal transposition without colostomy:functional results. Pediatr Surg Int 1999; 15:221-3.
6. Peña A. Posterior sagittal anorectoplasty: results in the management of 332 cases of anorectal malformations. Pediatr Surg Int 1988;3:94-104.
7. Okada A, Kamata S, Imura K, et al. Anterior sagittal anorectoplasty for rectovestibular and anovestibular fistula. J Pediatr Surg 1992;27:85-8.
8. Liu G, Yuan J, Geng J, et al. The treatment of high and intermediate anorectal malformations: one stage or three procedures? J Pediatr Surg 2004;39:1466-71.
9. Adeniran JO. One stage correction of imperforate anus and rectovestibular fistula in girls: preliminary results. J Pediatr Surg 2002; 37:E16.
10. Patwardhan N, Kiely EM, Drake DP. Colostomy for anorectal anomalies: high incidence of complications. J Pediatr Surg 2000; 36:795-8.
11. Peña A, Migotto-Krieger M, Levitt MA. Colostomy in anorectal malformations: a procedure with serious but preventable complications. J Pediatr Surg 2006;41:748-56.
12. Loulah MA, Sultan TA, Zeina WO. Primary versus multistage repair of congenital rectovestibular fistula. Menoufia Med J 2015; 28:813-7.
13. Mohammed K, Khalid S, Tarek AG, etal. One or two stage procedure for repair of rectovestibular fistula: which is safer? A single institution experience. Afr J Paediatr Surg 2017;14: 27-31.
14. Wakhlu A, Kureel SN, Tandon RK, et al. Long-term results of anterior sagittal anorectoplasty for the treatment of vestibular fistula. J Pediatr Surg 2009;44:1913-9.
15. Elsaied A, Aly K, Thabet W, et al. Two-stage repair of low anorectal malformations in girls: is it truly a setback? Ann Pediatr Surg 2013;9:69-73.
16. Menon P, Rao KL. Primary anorectoplasty in females with common anorectal malformations without colostomy. J Pediatr Surg 2007;42:1103-6.
17. Kumar K, Kandpal DK, Sharma SB, et al. Single-stage repair of vestibular and perineal fistulae without colostomy. J Pediatr Surg 2008;43:1848-52.
18. Gupta A, Agarwal S, Sreenivas V, et al. Primary definitive procedure versus conventional three-staged procedure for the management of low type anorectal malformation in female: a randomized controlled trial. J Indian Assoc Pediatr Surg 2017;22:87-91.
19. Tsukawa C, Nishijima E. Surgical repair of rectovestibular fistula with normal anus. J Pediatr Surg 1999;34:1703-5.
20. Upadhyaya VD, Gopal SC, Gupta DK, et al. Single stage repair of anovestibular fistula in neonate. Pediatr Surg Int 2007;23:737-40.
21. Kuijper CF, Aronson DC. Anterior or posterior sagittal ano-rectoplasty without colostomy for low-type anorectal malformation: how to get a better outcome? Pediatr Surg Int 2010;45:1505-8.
22. Rintala RJ, Lindahl HG, Rasanen M. Do children with repaired low anorectal malformations have normal bowel function? Pediatr Surg Int 1997;32:823-6.
23. Javid PJ, Barnhart DC, Hirschl RB, et al. Immediate and long term results of surgical management of low imperforate anus in girls. J Pediatr Surg 1998;33:198-203.
24. Hamid CH, Holland AJ, Martin HC. Long-term outcome of anorectal malformations: the patient perspective. Pediatr Surg Int 2007;23:97-102.

บทคัดย่อ **ผลการรักษาเด็กที่ไม่มีทวารหนักแต่กำเนิดชนิดของ Vestibular Fistula โดยไม่ทำ Colostomy**

สุจิตรา เก็บทอง, พบ, อัจฉริยา ทองสิน, พบ, วราภรณ์ มหธราดล, พบ.
กลุ่มงานศัลยศาสตร์ สถาบันสุขภาพเด็กแห่งชาติมหาราชินี กรุงเทพฯ

ความเป็นมา: ทารกเพศหญิงที่ไม่มีรูทวารหนักและมีรูเปิดอยู่หน้าต่อช่องคลอด สามารถที่จะรักษาโดยการผ่าตัด ทั้งชนิดผ่าตัดครั้งเดียวไม่มีการเปิดทวารเทียมหน้าท้องดักหน้าไว้ก่อน หรือจะผ่าตัดหลายครั้งเป็นขั้นตอนโดยมีการเปิดทวารเทียมป้องกันไว้ก่อน แบบที่เคยทำกันมาในอดีตก็ทำได้ การผ่าตัดให้สำเร็จในครั้งเดียวได้รับความนิยมมากขึ้นในปัจจุบันนี้

วัตถุประสงค์: การศึกษาครั้งนี้มีวัตถุประสงค์เพื่อประเมินผลลัพธ์ของการรักษาทารกที่ไม่มีรูทวารหนักแต่กำเนิดและมีรูเปิดหน้าต่อช่องคลอด ที่รักษาโดยการผ่าตัดครั้งเดียวโดยไม่ได้เปิดทวารเทียมที่หน้าท้อง ระหว่างปี พ.ศ. 2555 ถึง พ.ศ. 2559 ที่สถาบันสุขภาพเด็กแห่งชาติมหาราชินี ข้อมูลทั่วไป ชนิดหรือวิธีการผ่าตัด และผลลัพธ์ของการผ่าตัด ถูกนำมาตรวจสอบ

ผลการศึกษา: ในระยะที่ทำการศึกษา ทารกเพศหญิง 34 ราย ที่ไม่มีรูเปิดของทวารหนัก และมีรูเปิดที่หน้าช่องคลอดได้รับการรักษาชนิดผ่าตัดครั้งเดียวโดยไม่เปิดทวารเทียมที่หน้าท้อง ที่สถาบันของเรา รูเปิดที่อยู่หน้าต่อช่องคลอด (vestibular fistula -VF) ถูกจัดออกเป็น 2 ชนิด คือ รูเปิดที่ชี้ไปทางทวารหนักด้านล่าง (anovestibular fistula - AVF) จำนวน 15 ราย (ร้อยละ 44.1) และ รูเปิดที่ชี้ขึ้นไปหาไส้ตรงด้านบน (rectovestibular fistula- RVF) จำนวน 19 ราย (ร้อยละ 55.9) ผู้ป่วยส่วนใหญ่คลอดครบกำหนดและมีน้ำหนักแรกเกิดปกติ ความผิดปกติของหัวใจและหลอดเลือด เป็นความพิการแต่กำเนิดที่พบบ่อยที่สุด ทารกที่มี AVF ทำการผ่าตัด cut-back anoplasty (5 ราย) anal transfer (4 ราย) และ anterior sagittal anorectoplasty หรือ ASARP (6 ราย) ทารกที่มี RVF ทำผ่าตัดโดยวิธี anal transfer (5 ราย) และ ASARP (14 ราย) แผลติดเชื้อและแผลผ่าตัดแยกเป็นภาวะแทรกซ้อนที่พบได้บ่อยที่สุดหลังการผ่าตัด พบประมาณร้อยละ 15 และร้อยละ 25 ตามลำดับ การหดกลับของทวารที่สร้างใหม่และการเกิดซ้ำของรูเปิดหน้าต่อช่องคลอดพบได้อย่างละ 1 ราย ซึ่งภาวะแทรกซ้อนทั้งสองชนิดนี้ จำเป็นต้องมีการผ่าตัดแก้ไขซ้ำในหลายสัปดาห์ต่อมา แต่ก็ไม่ได้เปิดทวารเทียมที่หน้าท้องป้องกันแต่อย่างใด หนึ่งในสามของผู้ป่วยทั้งหมด มีอาการท้องผูก ซึ่งเกิดกับผู้ป่วยที่มี RVF บ่อยกว่า AVF อาการท้องผูกรักษาโดยการสอนเรื่องฝึกการขับถ่ายอุจจาระ รับประทานยาระบาย และสวนอุจจาระในบางครั้ง ไม่มีการเสียชีวิตในผู้ป่วยทั้งหมดที่ศึกษาครั้งนี้

สรุปผลการศึกษา: ทารกเพศหญิงที่ไม่มีรูเปิดของทวารหนัก และมีรูเปิดอยู่หน้าต่อช่องคลอด สามารถรักษาได้อย่างปลอดภัยด้วยวิธีการผ่าตัดครั้งเดียว โดยไม่ได้เปิดทวารเทียมที่หน้าท้อง เพื่อป้องกันไว้ก่อน จากการศึกษาครั้งนี้แม้ว่าจะมีภาวะแทรกซ้อนที่สำคัญ จำเป็นต้องผ่าตัดแก้ไขซ้ำ ผู้ป่วยก็สามารถรักษาโดยการผ่าตัดแก้ไขใหม่ได้สำเร็จและไม่ต้องเปิดทวารเทียมที่หน้าท้อง