Pediatric Surgery International (2019) 35:1109–1114
https://doi.org/10.1007/s00383-019-04540-y

ORIGINAL ARTICLE

The change over time in the postoperative bowel function in male anorectal malformation patients who underwent sacroperineal anorectoplasty and sacroabdominoperineal anorectoplasty

Toshio Harumatsu[1] · Masakazu Murakami[1] · Keisuke Yano[1] · Shun Onishi[1] · Koji Yamada[1] · Waka Yamada[1,2] · Ryuta Masuya[1] · Takafumi Kawano[1] · Seiro Machigashira[1] · Kazuhiko Nakame[1] · Motoi Mukai[1] · Tatsuru Kaji[1,2] · Satoshi Ieiri[1]

Accepted: 1 August 2019 / Published online: 7 August 2019

Abstract

Purpose Many kinds of operative procedures have been proposed for anorectal malformation (ARM) patients. At our institution, sacroperineal or sacroabdominoperineal anorectoplasty (SP-SAP) have been performed from 1984 to 2007. The aim of this study is clarify the change over the time in the postoperative bowel function in male ARM patients.

Methods Patient data were collected from 1984 to 2007. Fifty-two male patients with high- and intermediate-type ARM were enrolled. The patients' characteristics and bowel function were reviewed and analyzed retrospectively. The bowel function was evaluated according to the evacuation score (ES) of the Japan Society of ARM Study Group.

Results The operative procedures were SP-SAP in 52 male patients. The total ES improved chronologically and significantly until 11 years of age. Regarding the clinical stratification of the ES, the ratio of "excellent" and "good" results was over 91.9% at 11 years of age. A satisfactory bowel movement score was achieved by 9 years of age. The constipation, incontinence and soiling scores improved slowly but continuously until 11 years of age.

Conclusion The ES showed continuous improvement after a definitive operation. An understanding of the characteristics of improvement is very important in managing the postoperative bowel function in ARM patients.

Keywords Anorectal malformation · Long-term outcome · Bowel function · Sacroperineal anorectoplasty · Sacroabdominoperineal anorectoplasty

Introduction

Anorectal malformations (ARMs) are common congenital anomalies with an incidence of 1 in 5000 and associated with a number of issues, such as fecal continence, after definitive anorectoplasty. The first case of anorectoplasty for ARM was reported by Rhoads with an abdominoperineal approach in 1948 [1]. The sacroabdominoperineal (SAP) approach was later reported by Stephens [2]. In 1982, posterior sagittal anorectoplasty was reported by de Vries and Peña [3].

Many kinds of operative procedures have been proposed for ARM patients over the years. In our institution, the high and intermediate ARMs have been treated by sacroperineal or sacroabdominoperineal (SP-SAP) rectoplasty for the past 20 years. Recently, posterior sagittal anorectoplasty (PSARP) and laparoscopy-assisted anorectoplasty was introduced [4].

Ensuring a good bowel function after anorectoplasty is the most important point for maintaining a high quality of life in patients with ARM. In general, the postoperative bowel function in ARM patients after definitive operation is thought to improve with age. However, detailed characteristics of the changes in the postoperative bowel function among patients treated with SP-SAP procedure have never been reported.

✉ Satoshi Ieiri
sieiri@m.kufm.kagoshima-u.ac.jp

[1] Department of Pediatric Surgery, Research Field in Medicine and Health Sciences, Medical and Dental Sciences Area, Research and Education Assembly, Kagoshima University, 8-35-1 Sakuragaoka, Kagoshima city 890-8520, Japan

[2] Clinical Training Center, Kagoshima University Hospital, Kagoshima, Japan

The aim of this study was to evaluate the change over time in the long-term postoperative bowel function of male high- and intermediate-type ARM patients after definitive anorectoplasty performed by SP-SAP procedure.

Materials and methods

We conducted a retrospective review of all patients who underwent definitive surgery at our institution from 1984 to 2007. Fifty-two male patients with high- and intermediate-type ARM who underwent a definitive operation at our institution were enrolled. All of the ARM patients were followed for at least 3 years after anorectoplasty and closure of the colostomy. All of the patients have been followed regularly since birth. The patients' characteristics, type of ARM, associated anomalies, postoperative complications and bowel function were reviewed based on their medical records and analyzed retrospectively. The bowel function was evaluated according to the evacuation score (ES) of the Japan Society of Anorectal Malformation Study Group [5].

This study was performed in accordance with the Ethical Guidelines for Medical and Health Research Involving Human Subjects by the Ministry of Health, Labour, and Welfare of Japan in 2014. The study complied with the 1964 Declaration of Helsinki (revised in 2013) and was approved by the local ethics committee of our institution (registration number: 27-119). All participants or their parents provided their informed consent for involvement in this study.

Statistical analyses were performed using the repeated measures ANOVA. Probability values of less than 0.05 were considered to be statistically significant. Data are expressed as the mean ± standard deviation.

Operative procedure

All patients underwent colostomy as newborns. Before anorectoplasty, colostography and urography were performed to evaluate the type of ARM. The operative procedures were SP-SAP during the period between 1984 and 2007 at our institution. After anorectoplasty had been performed, the urethral catheter was removed on the seventh postoperative day with the confirmation of no leakage from the urethra by distal colostogram and urography. Dilation of the neo-anus was started on the 14th postoperative day. The colostomy was closed about 3 months after the definitive operation.

SP-SAP procedure

The SP procedure was performed for intermediate-type ARM. Two skin incisions were made at the sacroperineal region (sagittal incision from the sacrum to the perineum and neo-anus). Transection of the fistula was performed through the sagittal incision without laparotomy. After confirming the puborectal sling through the sacroperineal incision, the transected rectum was pulled through toward the neo-anus.

The SAP procedure was performed for high-type ARM. The principles of this technique were described by Stephens [2]. Two skin incisions (similarly to the SP procedure) were made and laparotomy was performed. Transection of the fistula was performed by an abdominal approach. Pull-through was performed in the same fashion as the SP procedure. The most important point of these procedures was to avoid injuring the muscle complex, including the puborectal sling and external sphincter muscle.

The evaluation of the postoperative bowel function

The follow-up data for patients older than 3 years of age were analyzed retrospectively in high- and intermediate-type ARM male patients. The bowel function was evaluated at 3, 5, 7, 9, and 11 years of age using the ES of the Japan Society of Anorectal Malformation Study Group [5]. The scores were based on four parameters: (1) frequency of bowel movement, (2) constipation, (3) incontinence, and (4) soiling. The total scores were analyzed with the sum of the frequency of the bowel movement score and soiling score. For constipation and incontinence, a lower score is preferred. The maximum score is 8 points, which indicates an excellent bowel function. The clinical stratification was evaluated according to the total ES as follows: 0–4, poor; 5–6, good; 7–8, excellent.

Results

Patient background data

The background data of patients are shown in Table 1. Fifty-two patients underwent the SP-SAP procedure for the following types of ARM: recto-prostatic urethral fistula in 2 (3.8%), recto-urethral fistula in 42 (80.8%), recto-bulbar fistula in 5 (9.6%), and anal agenesis without fistula in 3 (5.8%). Associated anomalies were as follows: esophageal atresia in 5 (9.6%), duodenal atresia in 2 (3.8%), urogenital in 24 (46.2%), cardiovascular in 7 (13.5%), abnormal sacrum in 7 (13.5%), and chromosomal in 4 (7.7%; all trisomy 21).

Clinical data

Table 2 shows the clinical data of ARM patients who underwent SP-SAP. The average age at anorectoplasty was 5.19 ± 1.71 months, and the body weight was 6.88 ± 1.06 kg. The operative time was 195.83 ± 130.71 min and the blood

Table 1 Background of patients

Number of patients	52
Lesion type	
Recto-prostatic urethral fistula	2 (3.8%)
Recto-urethral fistula	42 (80.8%)
Recto-bulbar fistula	5 (9.6%)
Anal agenesis without fistula	3 (5.8%)
Associated anomalies	
Esophageal atresia	5 (9.6%)
Duodenal atresia	2 (3.8%)
Urogenital	24 (46.2%)
Cardiovascular	7 (13.5%)
Abnormal sacrum	7 (13.5%)
Chromosomal (trisomy 21)	4 (7.7%)

Table 2 Clinical data

Body weight at operation (kg)	6.88 ± 1.06
Age at operation (months)	5.19 ± 1.71
Operative time (min)	195.83 ± 130.71
Blood loss per body weight during operation (ml/kg)	3.78 ± 2.40
Postoperative hospital stay (days)	22.9 ± 7.75

Table 3 Postoperative complications

Rectal prolapse	15 (28.8%)
Operation	11 (21.2%)
Age at operation	5.8 years
Rectal stenosis	9 (17.3%)
Operation	6 (11.5%)
Age at operation	9.5 years
Surgical site infection	2 (3.8%)

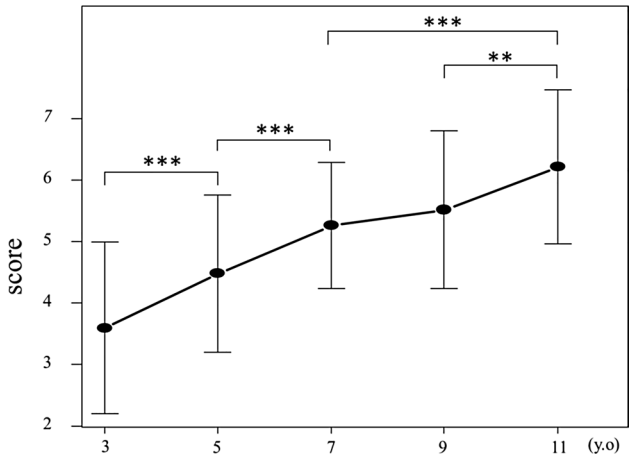

Fig. 1 The change over the time in the total evacuation scores. $^{**}p < 0.01$, $^{***}p < 0.005$

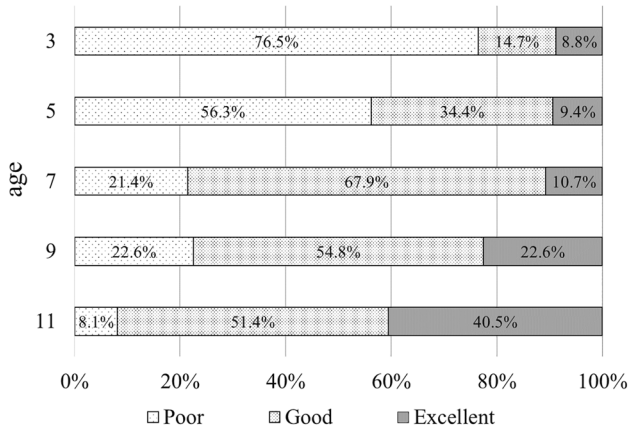

Fig. 2 The proportion of total evacuation scores for each age

loss per body weight during operation was 3.78 ± 2.40 ml/g. The postoperative hospital stay was 22.9 ± 7.75 days.

The operative complications are shown in Table 3. Fifteen patients (28.8%) had rectal prolapse, and 11 of them required operation. The mean age at anorectal prolapse was 5.8 years. Nine patients (17.3%) had rectal stenosis, and 6 of them required operation. The mean age at rectal stenosis was 9.5 years. Two patients (3.8%) had surgical site infections that were treated conservatively.

The postoperative bowel function

Figure 1 shows the total ES in each age. The mean total ES for each age was as follow: 3 years old; 3.82 ± 1.56, 5 years old; 4.54 ± 1.39, 7 years old; 5.17 ± 1.07, 9 years old; 5.50 ± 1.36, and 11 years old; 5.97 ± 1.16. The total ES improved chronologically and significantly until 11 years of age, and patients achieved satisfactory results at least 10 years after definitive operation. Figure 2 shows the proportions of the total evacuation scores for each age. The proportion of each clinical stratification for each age was as follows: 3 years (excellent 8.8%, good 14.7%, poor% 76.5), 5 years (excellent 9.4%, good 34.4%, poor 56.3%), 7 years (excellent 10.7%, good 67.9%, poor 21.4%), 9 years (excellent 22.6%, good 54.8%, poor 22.6%), 11 years (excellent 40.5%, good 51.4%, poor 8.1%).

The detailed scores and the changes in the frequency of bowel movement, constipation, incontinence, and soiling scores over the time are shown in Fig. 3. Regarding bowel movement, a satisfactory score was achieved by 9 years of age (Fig. 3a). The constipation, incontinence and soiling scores improved slowly until 11 years of age (Fig. 3b–d).

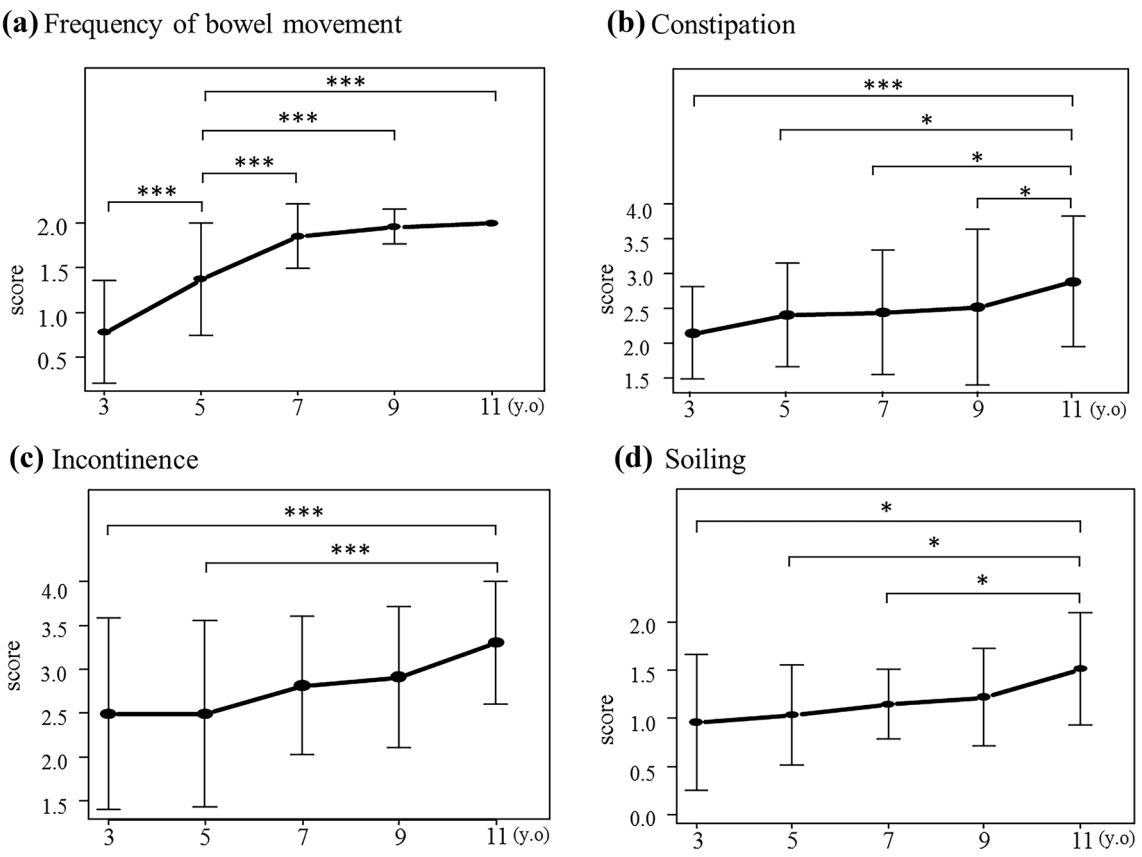

Fig. 3 The detailed scores and the changes over the time. **a** Frequency of bowel movement score. **b** Constipation score. **c** Incontinence score. **d** Soiling score. *$p < 0.05$, ***$p < 0.005$

Discussion

In the present study, we retrospectively reviewed our-single institution's experience with ARM in male patients and evaluated the changes over the time in the postoperative bowel function of patients who underwent SP-SAP. The major findings of this study were as follows: (1) 52 patients underwent SP-SAP between 1984 and 2007. (2) The total ES improved chronologically and significantly until 11 years of age. (3) The clinical stratification of ES also improved chronologically and significantly, and the ratio of "excellent" and "good" was over 90%. (4) A satisfactory bowel movement score was achieved by 9 years of age. (5) The constipation, incontinence and soiling scores improved slowly but significantly until 11 years of age.

Classical anorectoplasty procedures, such as abdominoperineal, SP and SAP, were performed before the PSARP procedure was introduced. Regarding the postoperative bowel function, Rintala compared the function achieved with SP-SAP with that achieved with PSARP [6], finding that PSARP was clearly superior to the SP-SAP procedure in terms of the bowel function and fecal continence. The internal anal sphincter function could be detected manometrically (anorectal reflex) in 76% of PSARP patients but were not detected in SP-SAP patients. Unfortunately, in the present study, a postoperative manometric study was not performed. However, the SP-SAP group showed gradual improvement of the bowel function, especially regarding the constipation, incontinence, and soiling score. With the SP-SAP procedure, the whole muscle complex structure was difficult to detect from the incision. Due to the above reason, this procedure could not pull-through the center of the muscle complex in the correct manner.

de Vries and Peña first reported PSARP in 1982, and since then, this procedure has come to be performed around the world. PSARP has gained overall acceptance as a standard operative method for repairing high and intermediate ARM. The PSARP procedure is still being modified, and as such, evaluations of its efficacy are still being performed [7, 8]. Peña himself reported that 37.5% of patients who underwent PSARP achieved a normal bowel function [9]. However, in some reports, < 15% of patients who underwent PSARP retained a normal bowel function during long-term follow-up [10–12]. Peña also reported that these patients require the postoperative bowel management for fecal incontinence [9]. Ninety percent of patients who underwent PSARP ultimately

achieved a satisfactory bowel function with appropriate postoperative bowel management [9]. In our report, the total ES improved chronologically and the clinical stratification of ES also improved until 11 years of age. Regarding the clinical stratification, at 11 years of age, the ratio of "good" results was 51.4%, while the ratio of "excellent" results was 40.5%. Our report showed that 91.9% of patients who underwent SP-SAP achieved a satisfactory bowel function. However, at 3 years of age, the bowel function was classified as "poor" in 76.5% of cases. The continuous improvement of these bowel function scores came from the growth of the patient and continuous bowel management, which was provided for a long time after the procedure. The SP-SAP procedure might be inferior to the PSARP procedure; however, continuous postoperative bowel management was important for achieving a good bowel function.

The age at operation for anorectoplasty may have affected the bowel function. Kuroda reported the development of the sphincter muscle using animal experiments and human samples obtained from ARM patients [13]. He examined the characteristics of the muscle fibers and clarified the type of muscle fibers as well. In patients with high ARMs younger than 6 months of age, type II fibers were dominant, whereas in older patients, type I fibers were increased, as in the normal population. This means that the function of the fibers of the sphincter muscle is established at 6 months old. One speculation is that definitive operation before the establishment of the sphincter muscle function may aid in obtaining a favorable postoperative bowel function. In our study, the mean age at anorectoplasty of SP-SAP was 5.2 months. The details of the mechanism underlying the slow recovery of the bowel function was unclear at present, although the age at definitive operation may be involved. Rintala reported that constipation disappeared at adolescence, and this was associated with an improved fecal continence outcome. This was clearly related to the growth and sexual maturation experienced during puberty [14].

Several limitations associated with the present study warrant mention. First, we evaluated the bowel function using a domestic scoring system, so we cannot compare our findings with those of other international reports. Second, the number of patients was small, and we only evaluated the bowel function. We must continue to perform long-term follow-up, including that of the genitourinary function and social performance, in older patients.

Conclusion

The ES of SP-SAP showed continuous improvement after the definitive operation. The change over time in the improvement of the bowel function may be caused by the operative procedure, the operative age and the continuous postoperative bowel management. It is very important to manage the postoperative bowel function in ARM patients by understanding the characteristics of their improvement.

Acknowledgements We thank Mr. Brain Quinn for his comments and help with the manuscript. This study was supported by a Grant-in-Aid for Scientific Research from the Japan Society for the Promotion of Science (JSPS: 19K10485, 19K09150, 19K09078, 19K03084, 19K18061, 19K17304, 19K18032, 18K08578, 18K16262 17K10555, 17K11514, 17K10183, 17K11515, 16K10466, 16K10094, 16K10095, 16K10434, 16H07090).

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

References

1. Rhoads JE, Pipes RL, Randall JP (1948) A simultaneous abdominal and perineal approach in operations for imperforate anus with atresia of the rectum and rectosigmoid. Ann Surg 127(3):552–556
2. Stephens FD (1953) Imperforate rectum; a new surgical technique. Med J Aust 1(6):202–203
3. Pena A, Devries PA (1982) Posterior sagittal anorectoplasty: important technical considerations and new applications. J Pediatr Surg 17(6):796–811
4. Georgeson KE, Inge TH, Albanese CT (2000) Laparoscopically assisted anorectal pull-through for high imperforate anus–a new technique. J Pediatr Surg 35(6):927–930
5. Onishi S, Nakame K, Yamada K, Yamada W, Kawano T, Mukai M, Kaji T, Ieiri S (2016) Long-term outcome of bowel function for 110 consecutive cases of Hirschsprung's disease: comparison of the abdominal approach with transanal approach more than 30 years in a single institution - is the transanal approach truly beneficial for bowel function? J Pediatr Surg 51(12):2010–2014
6. Rintala RJ, Lindahl HG (1999) Posterior sagittal anorectoplasty is superior to sacroperineal-sacroabdominoperineal pull-through: a long-term follow-up study in boys with high anorectal anomalies. J Pediatr Surg 34(2):334–337
7. Zaiem M, Zaiem F (2017) Muscle complex saving posterior sagittal anorectoplasty. J Pediatr Surg 52(5):889–892
8. Tang S, Dong N, Tong Q, Wang Y, Mao Y (2007) Laparoscopic assisted endorectal pull-through with posterior sagittal approach to the repair of postoperative rectourethral and rectovaginal fistula. Pediatr Surg Int 23(11):1077–1080
9. Pena A, Hong A (2000) Advances in the management of anorectal malformations. Am J Surg 180(5):370–376
10. Rintala R, Mildh L, Lindahl H (1994) Fecal continence and quality of life for adult patients with an operated high or intermediate anorectal malformation. J Pediatr Surg 29(6):777–780
11. Hassink EA, Rieu PN, C RS (1993) Are adults content or continent after repair for high anal atresia? A long-term follow-up study in patients 18 years of age and older. Ann Surg 218(2):196–200
12. Nixon HH, Puri P (1977) The results of treatment of anorectal anomalies: a thirteen to twenty year follow-up. J Pediatr Surg 12(1):27–37

13. Kuroda T (1991) A study about the external anal sphincter function in patients with anorectal malformation from the viewpoint of defecation control. Keio Igaku 68(1):127–141
14. Rintala RJ, Lindahl HG (2001) Fecal continence in patients having undergone posterior sagittal anorectoplasty procedure for a high anorectal malformation improves at adolescence, as constipation disappears. J Pediatr Surg 36(8):1218–1221