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Original Article

Feasibility of One-stage Correction for Recto-bulbar Anorectal Malformations

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Abstract ***Background:*** The feasibility of one-stage correction for recto-bulbar anorectal malformations is unclear. We examined the complications and short-term outcomes of one-stage correction for recto-bulbar anorectal malformations in neonates. ***Methods:*** We retrospectively examined six neonates who underwent one-stage correction for recto-bulbar anorectal malformations between 2004 and 2011. We reviewed the postoperative complications and functional evaluations-via the Kelly score, Krickenbeck classification, and magnetic resonance imaging. ***Results:*** During the study period, nine neonates were diagnosed with recto-bulbar anorectal malformations. Among these, one-stage correction was performed in six patients. The recto-bulbar urethral fistulas were confirmed by urethrography. All cases underwent posterior sagittal anorectoplasty. The average Kelly score was 4.33±1.21. Using the Krickenbeck classification, one child had grade 3 constipation. Grade 3 soiling was not noted. One child had malpositioning of the rectum that was revealed by magnetic resonance imaging. ***Conclusions:*** One-stage correction for intermediate imperforate anus is feasible even during the neonatal period.

Key words One-stage correction; Kelly score; Krickenbeck classification; Posterior sagittal anorectoplasty; Recto-bulbar anorectal malformations

Introduction

Bell in 1787 was the first to treat imperforate anus by dissecting through the perineum and searching for the rectal ampulla, and the first successful operation by this method was performed by Campbell in 1790. However, these procedures made no allowance for the preservation of a functioning anus, and Rouz in 1851 was the first to point out the importance of preserving the sphincter fibers while dissecting through the perineum.[1] Although most surgeons preferred one-stage correction at that time, one-stage rectoplasty was reportedly associated with high morbidity and mortality. Stephens and Smith subsequently supported the use of a staged program of preliminary colostomy and later rectoplasty after 12-15 months,[2] and deVries and Peña reported posterior sagittal anorectoplasty (PSARP) as a new technique in 1982, for patients who had undergone previous colostomy.[3] The standard surgical correction for intermediate or high imperforate anus currently involves a three-stage repair;[4] colostomy is performed first, followed by PSARP or sacro-perineal anoplasty (SP), and finally stoma closure. Although there have been a few reports of one-stage repair,[5,6] patient outcomes remain unclear. We performed one-stage correction for recto-bulbar anorectal malformations and examined the complications and outcomes in neonates to evaluate its usefulness.

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Methods

Patients and Indication for One-stage Correction

Patients diagnosed with recto-bulbar anorectal malformations between 2004 and 2011 were reviewed. Patients with birth weights <2000 g were excluded from the study because they underwent three-stage repair. Patients with chromosomal abnormalities were also excluded from the study because of the possibility that the associated abnormalities (e.g., retardation) might affect the postoperative faecal evaluation.

One-stage Correction Procedure

Urethrography was used to confirm the presence of a fistula preoperatively. PSARP was performed as described by deVries and Peña.[3] Briefly, the skin was opened through a midsagittal incision and the parasagittal fibers and muscle complex were divided. After all the muscle structures had been divided, the posterior rectal wall was exposed. The bowel was then opened vertically to confirm the fistula and the rectum was separated from the urethra by dissection above the fistula. The fistula was closed by continuous sutures. The rectum was fully mobilised and placed in front of the levator muscle, within the muscle complex and external sphincter. The divided muscles were repaired using absorbable sutures. Anoplasty was performed with 12 interrupted absorbable sutures. The anatomical route was identified using an electrical stimulation device. PSARP was performed using a standard technique, with no special procedures necessary. After operation, an 18-Fr catheter was left in the anus for 7 days and intravenous antibiotics were administered for 48 hours. Twice-daily anal dilation was begun 2 weeks after the operation. All operations were performed in the same institution, by the same surgeon who had more than 10 years of clinical experience.

Outcome Evaluations

We reviewed the perioperative complications and functional evaluations in the six patients at 5-6 years of age. Functional evaluations were made using the Kelly score,[7] Krickenbeck classification,[8] and magnetic resonance imaging (MRI). The Kelly score is based on continence, soiling, and sphincter squeeze. Each criterion was classified on a scale of 0-2, with a possible maximum of 6 points. The Krickenbeck classification system is composed of three elements: a diagnostic category, a surgical procedure category, and a category documenting functional criteria (voluntary bowel movements, soiling, and constipation). All evaluations were performed by a pediatric surgeon on an outpatient basis. MRI was performed to look for sacral anomalies, with an anatomical evaluation done at that time. Malpositioning of the rectum was evaluated at the level of the ischial line (I-line) and M-line (midline between the pubococcygeal line and the I-line). Malpositioning was defined as the thickness on one side being more than twice that on the other side.[9]

Results

During the study period, nine patients were diagnosed with recto-bulbar anorectal malformations at our hospital, among whom three were excluded from the study because of low birth weight (<2000 g) (n=2) or chromosomal abnormality (n=1). Six patients therefore met the criteria for inclusion in the study. These six cases were consecutive apart from the excluded cases. There were no associated malformations in any case. All fistulas were confirmed by urethrography, wherein a 6-Fr catheter was inserted into the urethra and the fistula was identified (Figure 1).

The patient characteristics are summarised in Table 1. The mean birth weight was 2839±499 g (range, 2180-3364 g) and the mean birth week was 37.4±1.6 weeks

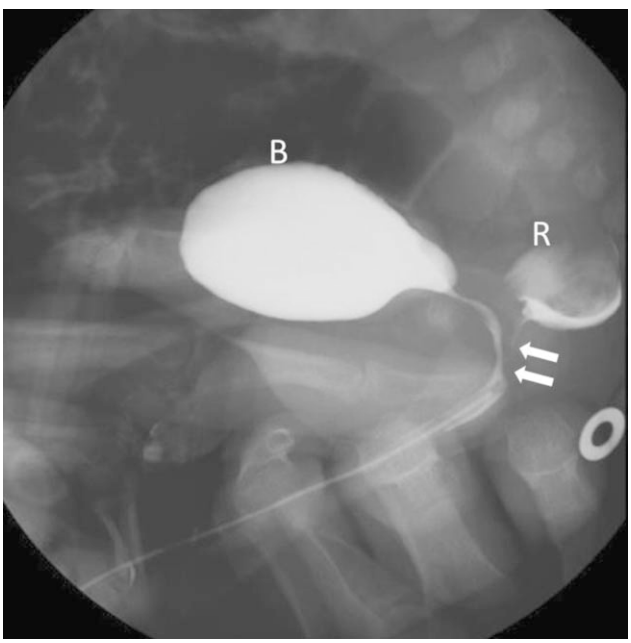

Figure 1 Fistulas were confirmed in all cases by urethrography. The fistula (arrow) connected to the rectum and urethral bulbar can be confirmed in this image. B: bladder; R: rectum.

(range, 36 weeks 0 days-40 weeks 4 days). The mean age at operation was 1.1 days (range, 1-2 days) and the mean operative time was 102±16 min (range, 76-118 min). There were no operative deaths or complications, except for an intraoperative bladder injury in one patient. However, this patient recovered with no complications, such as stenosis or fistula. No infectious complications were observed. The mean postoperative hospitalisation time was 43±18 days (range, 23-73 days). There were no early postoperative complications. There was no extension of hospitalisation due to surgical complication. The mean follow-up period in all six patients was 85 months (range, 62-108 months). The mean Kelly score at 5-6 years of age was 4.33±1.21 (mean continence 1.50±0.55, mean soiling 1.67±0.51, and mean sphincter squeeze 1.17±0.41) (Table 2). The results of the Krickenbeck classification are shown in Table 3. One child had grade 3 constipation. No patients showed grade 3 soiling. All patients underwent MRI postoperatively, before discharge, while still ≤2 months of age. Anatomical malpositioning was revealed in patient B (Figure 2). No sacral anomalies were identified.

Discussion

To the best of our knowledge, this study is the first to report on anus function following one-stage correction for recto-bulbar anorectal malformations in neonates. Traditionally, surgical correction of intermediate or high imperforate anus involves three-stage repair. After colostomy, it is recommended that definitive repair be performed at 2-3 months old.[4] Early operation has advantages including less time with a stoma, easier anal dilation, and no recognised psychologic sequelae from painful perineal maneuvers. In addition, the advantages of performing a single radical operation in the neonatal period include a single operation, avoiding a colostomy, and reducing wound breakdown.

Adeniran et al reported that one-stage correction was safe and that colostomy may be unnecessary in many patients with anorectal malformations.[5] Albanese et al also reported that postoperative stenosis occurred in one of five patients after one-stage correction, and there were no cases of perineal wound complications.[6] Similarly, there were no infectious complications in the current study. Bacterial colonisation of the bowel takes 1 week,[6] after which infections are much more likely. One of our patients experienced an intraoperative bladder injury. Hong et al reported that the risk of urologic injury was increased in patients undergoing repair without distal colostography.[10] Although all patients in the current study underwent preoperative urethrography, bladder injury could still occur. It is necessary to be particularly cautious because of the relatively shorter distance of the bladder from the urethra in neonates than in infants. The abrasion range is accordingly shortened, and rectal mobilisation is easier with one-stage than with three-stage repair. One-stage

Table 1 Patients' backgrounds

Factors	Value
Male: Female	6 : 0
Birth week	37.4±1.6 week (36 weeks 0 day - 40 weeks 4 days)
Birth weight (g)	2839± 499 (2180-3364)
Age at operation (days)	1.1 (1-2)
Operating time (minutes)	102±16 (76-118)
Postoperative complications	1 / 6 (1 : urethra injury, no infection)
Follow up periods (months)	85±15 (62-108)
Kelly score at 5 years old	4.33±1.21 (3-6)

Table 2 Kelly score results

Patient	Continence	Soiling	Sphincter squeeze	Total score	Follow-up (months)
A	1	1	1	3	87
B	1	1	1	3	108
C	2	2	1	5	93
D	1	2	1	4	87
E	2	2	2	6	75
F	2	2	1	5	62
Mean	1.50± 0.55	1.67 ±0.51	1.17± 0.41	4.33±1.21	85

correction is therefore no more difficult even during the newborn period, and can be performed safely, as reported previously.[5,6]

Fukata et al reported that the average Kelly score for patients with intermediate imperforate anus was 4.2,[11] while Senel et al reported an average Kelly score of 5.5 in patients with intermediate imperforate anus with rectobulbar urethral fistula.[12] The average score in our study was 4.33±1.21. Using the Krickenbeck classification, Hassett et al reported grade 3 constipation was noted in 42% of children, and grade 2 constipation was recorded in one child in the rectourethral group. One child in this group had grade 3 soiling.[13] Tong et al, using MRI to evaluate rectal malpositioning in a PSARP group, reported malpositioning rates of 14.3% at the I-line and 10.7% at the M-line.[9] The case of malposition shown by MRI had a worse result in both Kelly score and Krickenbeck classification compared with other cases. MRI findings and functional outcomes are considered to be correlated in our series. Correction in the neonatal period allows the possibility of developing neural networks and synapses,[14,15] and training of defecation from an early age can lead to improvements in defecation function.[6,16] However, there was no evidence of improved defecation function in our study, and contraction was weak around the anus, reflected by the sphincter squeeze aspect of the Kelly score and constipation of the Krickenbeck classification. PSARP in the neonatal period may be associated with a greater risk of scarring while dissecting the anal sphincter, because the anal sphincter is thin. Rintala et al compared PSARP and SP,[17,18] and reported that PSARP was more likely to cause constipation than SP. Constipation following PSARP is considered to be related to abnormal movement of the anal sphincter. The SP method is thus associated with the possibility of a good prognosis in the case of newborns.

This study was limited by the small number of cases and the relatively short follow-up period. Furthermore, there is currently no agreement on absolute parameters in anorectal anomalies.[19,20] We used the Kelly score, Krickenbeck classification, and MRI because they are commonly reported for outcome assessments.[13,20] There is currently no consensus opinion on what parameters to evaluate at each age, though soiling is difficult to determine in infants, and we therefore evaluated it only in subjects older than 5 years in this study.

Table 3 Krickenbeck classification results

Patient	Voluntary bowel movements	Soiling	Constipation
A	Yes	Grade 1	Grade 3
B	No	Grade 2	Grade 2
C	Yes	No	Grade 2
D	Yes	No	Grade 2
E	Yes	No	Grade 1
F	Yes	No	Grade 2

Conclusion

One-stage correction for recto-bulbar anorectal malformations is safe and feasible, even in the neonatal period. One-stage correction has several advantages including no need for colostomy, thus avoiding problems related to colostomy and reducing the burden on family life. However, further, longer-term observations and large-scale studies are required to confirm the suitability of this procedure.

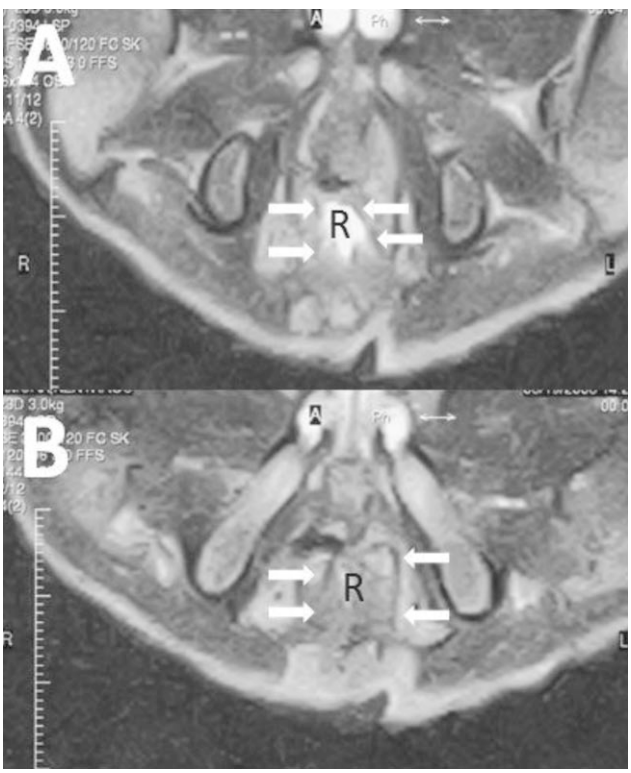

Figure 2 Postoperative axial T2-weighted magnetic resonance imaging of the pelvis. Muscles can be seen around the rectum. Normally they are symmetrical. Malpositioning of the rectum and muscle (arrows) with a right-left difference at the M-line (A) and I-line (B) in patient B. R: rectum.

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Declaration of Interest

The authors declare that they have no conflict of interests.

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