

INTERMITTENT ASSESSMENT OF PATIENTS WITH REPAIRED ANORECTAL MALFORMATIONS: RECOVERY OF BOWEL AND ANORECTAL FUNCTIONS WITH PATIENT AGE

OPERE EDİLMİŞ ANOREKTAL MALFORMASYONLU HASTALARIN BELİRLİ ARALIKLARLA DEĞERLENDİRİLMESİ: BARSAK VE ANOREKTAL FONKSİYONLARIN YAŞLA GELİŞİMİ

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Gazi Medical Journal 2003; 14:159-166

ABSTRACT

Purpose: The aim of this prospective study was to evaluate the relation between bowel functions and anorectal manometric assessment scores by comparing these 2 parameters with increasing patient age. **Methods:** A total of 13 patients whose anorectal malformations had been repaired were evaluated at least 2 to 4 times yearly between 1997 and 2003. Bowel functions were assessed according to a scoring system. The maximum bowel function score (BFS) was 14 points. Anal canal resting and squeeze pressures, rectoanal inhibitory reflex, and sense of defecation and urgency were determined manometrically, each corresponding to 1 point when present. The maximum manometric score (MS) was 5 points. The patients with constipation were treated with laxatives or enemas, the other patients who had soiling due to a short colon were treated with loperamide hydrochloride and one patient who had a failed operation was treated with a redo procedure. **Results:** After the particular treatment for each patient, both BFS and MS were raised, in general. The MS of patients increased significantly with the advancement of therapy, but reached a plateau and showed no progression thereafter. BFS, on the other hand, continued to increase with rising patient age. While the 2 scores were correlated at the initiation of therapy, they were not correlated with progression of therapy, with BFS surpassing MS. When the MS of the patients with a BFS under 7 points was compared with that of others over 7 points, no significant difference was found. **Conclusion:** Both BFS and MS increased after treatment, but they did not change with increasing patient age. Good bowel functions were not associated with good anorectal functions.

Key Words: Anorectal Malformation, Bowel Function, Manometry.

INTRODUCTION

Since the level of fecal continence directly

ÖZET

Amaç: Bu çalışmada barsak fonksiyonları ile birlikte anorektal monometrik ölçümler arasındaki ilişki hasta yaşı yönünden prospektif olarak karşılaştırılmıştır. **Metod:** 1997 ile 2003 yılları arasında toplam 13 ARM'lu çocuk en az iki, en fazla dört kez bir yıl aralıklarla değerlendirildi. Barsak fonksiyonları bir puanlama yöntemiyle değerlendirilerek en fazla barsak fonksiyon derecesi (BFD) 14 olarak belirlendi. Anorektal fonksiyonlar ise manometrik çalışma ile değerlendirildi. Anal kanal dinlenme (DB) ve sıkma (SB) basınçları, rektoanal inhibitör refleks (RAİR), defekasyon ve acil defekasyon hissi saptanarak, herbirinin varlığı 1 puan olarak kabul edildi ve en fazla anorektal fonksiyon derecesi (AFD) 5 olarak belirlendi. Takipte kabızlık yakınması olanlara laksatif ve lavman tedavisi, soiling yakınması olanlara başlıca loperamid-HCl tedavisi verildi. Bir hasta ise önceki başarısız ameliyat nedeniyle opere edildi. **Bulgular:** Genel olarak tüm çocukların AFD ve BFD değerleri uygulanan tedaviler sonrasında yükselmiş olarak bulundu. BFD düzeyi çocuğun yaşı ilerledikçe artmaya devam ederken, AFD'deki artışın bir noktadan sonra durakladığı saptandı. BFS'si 7'nin altında olan çocukların MS sonuçları, BFS'si 7'nin üstünde olanlarla kıyaslandığında anlamlı bir fark olmadığı gözlemlendi. Ayrıca, çocuğun klinik durumuyla AFD arasında bir korelasyon bulunmadı. **Sonuç:** Tedavi sonrasında hem BFS hem de AFD'de düzelme olmuştur. Ancak çocuğun yaşı ilerlemesine rağmen bu düzelmede bir değişiklik göze çarpmamıştır. İyi barsak fonksiyonu, iyi anorektal fonksiyonla birlikte.

Anahtar Kelimeler: Anorektal Malformasyon, Barsak Fonksiyonu, Manometri.

affects patients' quality of life after the repair of anorectal malformations (ARMs) (1), a large

number of authors have described their experiences to explain and address this issue. While some reports have focused on anorectal functions and their effects on continence (2-4), it has been accepted that most patients with ARM have abnormal bowel motility (5, 6). Questionnaire-based scoring systems, colonic transit time and manometric studies have been used to evaluate the bowel function of patients with repaired ARM (7-9). Although postoperative continence differs according to the surgical techniques used, patients with lower ARM have better long-term functional outcomes than those with intermediate and high types (9). The aim of this study was to evaluate the bowel and anorectal functions of patients whose intermediate or high malformations had been repaired and had received treatment for fecal incontinence. The progression of the bowel and anorectal functions of these patients were determined and compared in terms of the treatment.

PATIENTS AND METHODS

Thirteen patients (4 girls, 9 boys) repaired for ARM were evaluated annually in terms of bowel and anorectal functions between 1997 and 2003 at consequent ages. Three of them had been operated on at another center. Five patients were evaluated twice, 5 patients 3 times, and 3 patients 4 times. The mean age at the first evaluation was 7 years (3-12 years of age) and at the last one was

10.8 years (5-16 years of age). Table 1 shows the original malformations and initial corrective operations of the patients. It also shows the complaints of these patients when they sought treatment and our treatment protocols during the follow-up period. At the beginning of the evaluation of each patient, contrast enema images were studied after a careful abdominal and rectal examination to document the cause of the fecal incontinence. The patients with a short rectum and sigmoid colon had fecal soiling and frequent defecations in a day. These patients were generally treated with a constipating diet, regular sitting on the toilet, loperamide hydrochloride or sometimes evacuating saline enemas. The other patients with rectosigmoid dilatation had overflow soiling because of constipation and received a high fiber laxative diet and/or evacuating saline enemas. The agent most preferred as an aperient was lactulose.

Bowel functions were assessed using a scoring system modified by Rintala et al. (10). According to this, the maximum bowel function score (BFS) was 14 (Table 2). Half of the maximum BFS was accepted as the cut-off point and the patients with a BFS under 7 points were considered clinically weak. BFS was assessed by means of a questionnaire filled in by the parents or conscious patients (Table 2).

Anorectal functions were assessed by manometry. Manometric evaluations of the

Table-1: Types of ARM, initial operations, complaints and therapies of the patients.

Number of Patient	Types of Malformations	Initial Pull-through Operations	Complaints	Types of therapy during follow-up period
1	RPROS	SP	Soiling	Constipating diet, loperamide HCl
2	RVEST	P	Constipation	Laxative diet, enema
3	RPROS	SP	Soiling	Constipating diet, loperamide HCl
4	CLM	SP	Constipation	Laxative diet, aperients, enema
5	RBUL	SP	Soiling	Constipating diet, loperamide HCl
6	RBUL	SP	Soiling	Constipating diet
7	RVAG	SP	Constipation	Laxative diet, aperients
8	RBUL	PSARP	Soiling	Constipating diet, enema
9	RBUL	PSARP	Soiling	Toilet regularity
10	RPROS	SAP	Soiling	Toilet regularity
11*	RBUL	SP	Soiling	Redo-operation (PSARP)
12*	RBUL	SP	Soiling	Constipating diet, loperamide HCl, enema
13*	RVEST	P	Constipation	Laxative diet, aperients, enema, sigmoid resection

* Operated on at other centers.

(RPROS; rectourethral prostatic fistula, RVEST; rectovestibular fistula, CLM; cloacal malformation, RBUL; rectourethral bulbar fistula, RVAG; rectovaginal fistula, SP; sacroperineal, P; perineal, SAP; sacroabdominoperineal, PSARP; posterior sagittal anorectoplasty, RPROS: rectoprostic fistula, RVEST; rectovestibular fistula, CLM; cloacal malformation, RBUL; rectobulbar fistula, RVAG; rectovaginal fistula, SP; sacroperineal, P; perineal, SAP; sacroabdominoperineal, PSARP; posterior sagittal anorectoplasty).

Table-2: Assessment of Bowel Function.

Criteria	Score
Frequency of defecation	
Every other day-twice a day	2
More often	1
Less often	1
Urge sensation	
Always	3
Most of the time	2
Uncertain	1
Absent	0
Soiling	
Never	3
Staining less than 2/week, no change of underwear required	2
Frequent, once a day, require change of underwear	1
Constant	0
Constipation/Diarrhea	
Never	3
Manageable with diet	2
Manageable with medicin/enemas	1
Severe, not manageable	0
Social problems	
No problem	3
Sometimes (foul odor)	2
Restrictions in social life	1
Severe social and/or psychologic problems	0
The maximum score	14

patients were performed as described by Langemeijer-Molenaar (11) and Scharli (12). Resting pressure (RP) and squeeze pressure (SP) of the anal canal, rectoanal inhibitory reflex (RAIR), and defecation and urge senses were determined and 1 point was given for each when present. Accordingly, the maximum manometric score (MS) was 5 (Table 3).

Manometry technique:

Without any sedation, all studies were performed by the same technician experienced in anorectal manometry in the Motility Laboratory in the Gastroenterology Department of our hospital. All patients were cooperative during the procedure. The water-perfused catheter used in the study was 4.5 mm in diameter with 8 channels located circumferentially at 0.5 cm intervals and with a balloon at the tip. Mean RP over 25 mmHg of the anal canal was considered significant for scoring. An SP more than twice the RP was included in the scoring. The balloon was inflated with increasing volumes of air (10, 20, 30,...ml), and RAIR was considered to exist if there was a more than 25% decrease in RP. During the inflation of the balloon, the volumes corresponding to the first sense of defecation and the sense of urgency were determined.

The changes in BFS and MS before and after the treatment, the relation between BFS and MS

Table-3: Assessment of Anorectal Functions with Manometry.

Criteria	Score
Resting Pressure (RP) (> 25 mmHg)	1
Squeeze Pressure (SP) (> 2 x RP)	1
Rectoanal inhibitory reflex (RAIR)	1
Sense of defecation	1
Sense of urgency	1
The maximum score	5

and the improvements in BFS and MS with increasing patient age were evaluated at the end of the study period.

The other associated problems of the patients and their progress are shown in Table 4.

Statistical Analysis:

The correlation between the BFS and MS according to increasing patient age was analyzed with the Spearman rank correlation test. The MS of the patients with BFS under and over the cut-off points was compared using the Mann-Whitney U test. The Wilcoxon sign rank test was used to compare the results of BFS and MS at different ages. P less than 0.05 was considered statistically significant.

RESULTS

The most commonly found type of ARM was rectourethral bulbar fistula (6/13, 46%), followed by rectourethral prostatic fistula (3/13, 23%), rectovestibular fistula (2/13, 15%), rectovaginal fistula (1/13, 7.6%) and cloacal malformation (1/13, 7.6%). Most of the initial operations were sacroperineal pull-through (8/13, 61%), followed by posterior sagittal anorectoplasty (PSARP) (2/13, 15%), perineal pull-through (2/13, 15%) and sacroabdominoperineal pull-through (1/13, 7.6%).

We did not include the results of the fourth evaluations of BFS and MS in the statistical analyses due to the insufficient number of patients. The initial average BFSs and MSs were 5.1 and 2.7, respectively. BFS increased to 7.6 in the second evaluations and to 9.3 in the third ones. The second and third mean BFSs determined after the advent of patient-specific therapy were higher than the initial ones (p<0.05). The mean BFSs found in the second and third evaluations did not differ statistically, although the last one was higher than the second one (p>0.05). MS increased to 4 in the second

Table-4: Other associated problems of the patients (VUR: vesicoureteral reflux UTI: urinary tract infection).

Number of Patient	Other	Associated Problems
1		Right-sided grade 3 VUR (<i>regressed to grade 1</i>) Night time enuresis (<i>improved</i>)
2		Obesity (<i>under endocrinologic control</i>) Day and night time enuresis (<i>regressed to night time</i>) Recurrent UTI (<i>under antibiotic prophylaxis</i>)
3		Malrotation Bladder instability Day and night time enuresis (<i>regressed to night time</i>) Recurrent UTI (<i>improved</i>) Subcoronal hypospadias (<i>operated on</i>) Undescended testes (<i>operated on</i>)
4		Bilaterally grade 5 VUR (<i>reimplanted</i>) Neurogenic bladder (<i>bladder augmentation and Mitrofanoff procedure</i>) Bifid uterus, vaginal septum Tethered cord Scoliosis
5		Fusion between L-5 and S-1 vertebrae
6		Night time enuresis (<i>improved</i>) Fusion defect of S-1 vertebra
7		Left-sided grade 3 VUR (<i>resolved</i>) Day time enuresis (<i>improved</i>) Fusion defects of S-1 vertebrae Fascial asymmetry Deafness
8		Left paraureteral diverticulum (<i>operated</i>) Left-sided grade 3 VUR (<i>continued</i>)
9		None
10		Sacral hypoplasia Tethered cord
11		None
12		Low mental function Fusion defect of S-1 vertebra
13		None

Table-5: Results of the BFS and MS evaluations with increasing age (*BFS; bowel function score, MS; manometric score*).

	1st evaluation (n=13) *	2nd evaluation (n=13)	3rd evaluation (n=8)
BFS			
Mean ± SD	5.1 ± 3.7	7.6 ± 2.8	9.3 ± 3.4
Median (Min-Max)	4 (1-12)	8 (2-12)	9 (3-14)
MS			
Mean ± SD	2.7 ± 1.1	4 ± 0.9	3.8 ± 0.8
Median (Min-Max)	3 (1-5)	4 (2-5)	4 (3-5)

* p<0.05 versus second and third evaluations of the BFS and MS.

evaluation, but ceased to show progression, with a mean score of 3.8 in the third evaluation. Although the second and third mean MSs were

higher than the initial ones (p<0.05), the mean MSs found in the second and third evaluations did not differ statistically (p>0.05) (Table 5). A

Table-6: Comparison of MS results according to the BFS cut-off point.

	1st evaluation of MS (n=9)	2nd evaluation of MS (n=6)	3rd evaluation of MS (n=1)
BFS \leq 7 points			
Mean \pm SD	2.4 \pm 1	3.8 \pm 1.1	4
Median (Min-Max)	3 (1-4)	4 (2-5)	4
BFS \leq 8 points	(n=4)	(n=7)	(n=7)
Mean \pm SD	3.5 \pm 1.2	4.2 \pm 0.7	3.8 \pm 0.8
Median (Min-Max)	3.5 (2-5)	4 (3-5)	4 (3-5)

* p<0.05 versus second and third evaluations of the BFS and MS.

Table-7: Detailed evaluation results of BFS and MS according to increasing patient age (BFS; bowel function score, MS; manometric score).

Number of Patient	Age				BFS				MS			
	1	2	3	4	1	2	3	4	1	2	3	4
1	12	13	14	16	4	7	8	12	2	4	3	3
2	12	14			7	11			3	5		
3	7	9	13		2	6	14		3	2	3	
4	7	9	10	13	1	6	11	4	1	5	5	5
5	6	8	10	13	5	9	8	10	2	3	3	2
6	5	8			8	10			3	4		
7	6	8			4	8			2	5		
8	4	6	8		8	6	10		4	5	5	
9	4	5			12	10			5	5		
10	3	4	5		11	12	13		2	4	4	
11	10	11			2	2			4	4		
12	7	8	10		1	4	3		1	4	4	
13	8	9	11		2	8	8		3	5	4	

* p<0.05 versus second and third evaluations of the BFS and MS.

strong correlation between the BFS and MS was found in the first evaluation compared to the second and third ones ($r=0.5$ versus $r=0.1$ and $r=0.09$). According to these results, there was a significant improvement in both BFS and MS after the specific treatment. However, after the treatment, no significant recovery was found in terms of BFS or MS with increasing patient age.

The initial evaluation revealed 9 patients with a BFS equal to or less than 7 points. This number dropped to 6 patients in the second evaluation and to only 1 patient in the third. That patient had a rectourethral bulbar fistula corrected elsewhere. The initial evaluation revealed 4 patients with constipation and 5 with soiling. Four patients with soiling had a BFS above 7 points. This number increased to 7 patients in both the second and third evaluations. The MSs of the patients with a BFS above 7 points did not differ from those of the patients having BFSs less than 7 points, for all 3 evaluations ($p>0.05$) (Table 6). Therefore, good bowel functions were not found

to be correlated with good anorectal functions. Although almost all patients in this series showed good progress in terms of BFS, there was an undulation in 5 who discontinued the therapy proposed to them. However, the MSs of these 5 patients were not affected by therapy continuity (Table 7).

In some patients, the development of senses of defecation and urgency, and of RAIR increased the MS that had been low at the first evaluation. While the patients who had senses of defecation and urgency numbered 9 and 6, respectively, at the first evaluation, all patients had both senses at subsequent evaluations. The presence of RAIR increased from 5 patients at the first evaluation to 8 (4 constipation, 4 soiling) the next time. Changes in RP values because of an asymmetric and short anal canal caused a drop of 1 point in 3 patients during follow-up. SP values did not change for all evaluations.

Nine (69%) of 13 patients complained of

fecal soiling because of an insufficient rectosigmoid reservoir. In this group one patient's (no. 11) operation was unsuccessful and redo-PSARP was performed after the first evaluation. Although this patient did not have any sacral abnormality, his BFS did not change and there was a drop in his MS 6 months after the redo operation. Because he did not come to check ups for a long time, we do not know his current condition. While 7 of the other 8 patients with soiling had a rise in their BFS, 1 showed a drop. MSs were unchanged in 3 of these patients, increased in 4 and decreased in 1. One patient (no. 3) who had rectourethral prostatic fistula in this group had the highest BFS and is not receiving any treatment (Table 7).

Four (31%) of 13 patients suffered from constipation. Two of them benefitted from laxative treatment and had better BFSs and MSs. Although another patient in this group showed some improvement for both BFS and MS, she was operated on because of megasigmoid after the third evaluation and did not require laxative treatment after this operation. The last patient in this group (no. 4), who had cloacal malformation originally and severe constipation, showed good progress during the first 3 evaluations. However, because she discontinued the treatment we found a significant regression in her BFS, but not in her MS during the fourth evaluation (not used for statistical analysis) (Table 7). Since she also had a small and neurogenic bladder, sigmoid resection was performed for bladder augmentation, thus contributing to the constipation treatment. Now she has regular voluntary bowel movements with laxative therapy.

After the particular treatment for each patient in this series, improvements in BFSs and MSs were detected in 85% and 69% of patients, respectively. As increases in both BFSs and MSs were achieved in all constipated patients through treatment, this rate was only 30% in the patients with soiling. After the last check-ups of all patients, 7 of them continued with their protocols, 5 did not need any treatment and 1 was lost to follow-up.

Only 3 patients (23%) in this series were free of associated anomalies. Six patients (46%) had various sacral abnormalities. Two of them (15%) had a tethered cord and none resulted in cord

release. In addition, 5 patients (38%) had different types of urinary tract problems (Table 4).

DISCUSSION

When the patients with ARM are compared with normal peers, they have lower fecal continence rate in all types, especially for high malformations (9). In addition to the type of ARM, surgical techniques affect functional outcome. PSARP has a marked superiority to previous techniques in long-term results (13). The other important factors affecting future continence are the original rectum and sphincters, the length of the colon, the sacrum and the management of the patient after the definitive operation. In most patients in this series a sacral approach was used at first. These are the initial patients in our department. Since the other patients who underwent PSARP have not been evaluated sequentially yet, we are not able to present their results. The diversities of surgical techniques and surgeons may be a drawback of this study.

A relation between the manometric results of the anal canal and functional outcomes has been reported previously for intermediate and high ARM. Iwai et al. and Varma evaluated patients operated on before the PSARP era by using the Kelly score and anorectal manometry (3, 14, 15). They found a direct correlation between clinical and manometric assessment. Although Hedlund et al. found an incomplete correlation between them after PSARP (16), soiling was more common with low RP and low SP, and constipation was more common with large rectal volume. Rintala and Lindahl investigated patients who had undergone internal sphincter-saving PSARP for intermediate and high ARM and found a correlation between a good continence and the presence of a functional internal sphincter and high RP in their series (10). In our series, there was a strong correlation between the BFS and MS before treatment. This correlation weakened after treatment although both scores were higher. The improvement in anorectal and bowel functions after treatment did not vary with patient age during follow-up. On the other hand, better bowel functions were not compatible with better anorectal functions. Bowel functions were also found to be affected by therapy continuity more than by anorectal functions.

Preservation of the internal anal sphincter has been reported not to be essential but to be a contributing factor for the development of continence (4). Husberg et al. showed that a positive RAIR indicating internal sphincter function provided better anal continence to patients with high or intermediate ARM (17). In the current series, positive RAIR was determined in 8 patients at the end of the evaluation period. This positivity in 3 of 8 patients was absent at first and was then determined during follow-up. Positive RAIR 16 months after the definitive operation has also been reported by Varma (15). We sometimes had technical difficulties in detecting RAIR because of a short and asymmetric anal canal or weakness of the internal anal sphincter as well as RP values. According to our results, the main increase in MS was achieved by the development of rectal sense, followed by RAIR and SP. RP measurements were unreliable. Therefore, a second manometric assessment of the anal canal 1 year after the first assessment should give a more objective MS.

There is no consensus on the assessment of bowel function. Rintala proposed a detailed questionnaire to evaluate bowel function and suggested to compare these patients with a control group of healthy children of similar age and sex distribution (9). We used a questionnaire and tried to determine whether the parents or patients filled it in honestly or not by performing physical and rectal examinations during the check-ups. It was difficult to form a control group since one patient was investigated more than once at different times. However, we could compare the patients with themselves as they were growing up even though the number in this series was small.

In addition to the contribution of social adaptation of the patient, medical support and/or corrective surgery, and the course of the colonic motility may be determinative of fecal continence in adulthood. Holschneider et al. investigated rectal pouch and fistula specimens of 52 children with ARM and reported abnormal innervation patterns in 96% (18). While abnormal innervation of the rectum seemed to be related to the higher frequency of constipation in patients with low ARM, they could not find any relation between the specific neuronal abnormality and the clinical course of the patients with intermediate and high

ARM. In addition to investigations of the anorectum by different methods, such as manometry, ultrasonography (19), electrophysiology (20) and biopsy, some studies on the upper colonic segments exist. In one study, while there was rectosigmoid hypomotility in low ARM, right colonic motility was found to be prolonged in patients with high ARM (7). Heikenen et al. performed colonic manometry in patients with repaired ARM and complaining of constipation or incontinence (8). They found 81% of high amplitude propagating contractions originating from the proximal colon reached into the rectum of the patients, as normally propagating in 14%.

In conclusion, the present series once again showed the importance of follow-up after surgery and that appropriate treatment for each patient provided improvements in both bowel and anorectal functions. Although bowel and anorectal functions did not maintain their recovery with increasing patient age, bowel functions found susceptible to the therapy should be investigated with objective motility studies.

ACKNOWLEDGMENT

The authors thank Miss Ayten Erdem for her technical help with the manometry studies.

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REFERENCES

1. Bai Y, Yuan Z, Wang W, Zhao Y, Wang H. Quality of life for children with fecal incontinence after surgically corrected anorectal malformation. *J Pediatr Surg* 2000; 3: 462-464.
2. Arhan P, Faverdin C, Devroede G, Dubois F, Coupris L, Pellerin D. Manometric assessment of continence after surgery for imperforate anus. *J Pediatr Surg* 1976; 11: 157-166.

3. Iwai N, Ogita S, Kida M, Fujita Y, Majima S. A clinical and manometric correlation for assessment of postoperative continence in imperforate anus. *J Pediatr Surg* 1979; 14: 538-543.
4. Lin CL, Chen CC. Continence and rectoanal sphincteric inhibitory reflex after posterior sagittal anorectoplasty. *J Formos Med* 1996; 95: 303-307.
5. Peña A, El-Behery M. Megasigmoid: a source of pseudoincontinence in children with repaired anorectal malformations. *J Pediatr Surg* 1993; 28:199-203.
6. Peña A, Guardino K, Tovilla JM, Levitt MA, Rodriguez G, Torres R. Bowel management for fecal incontinence in patients with anorectal malformations. *J Pediatr Surg* 1998; 33: 133-137.
7. Rintala RJ, Marttinen E, Virkola K, Rasanen M, Baillie C, Lindahl H. Segmental colonic motility in patients with anorectal malformations. *J Pediatr Surg* 1997; 32: 453-456.
8. Heikenen JB, Werlin SL, Di Lorenzo C, Hyman PE, Cocjin J, Flores AF, Reddy SN. Colonic motility in children with repaired imperforate anus. *Digest Dis Sci* 1999; 44: 1288-1292.
9. Rintala RJ. Fecal incontinence in anorectal malformations, neuropathy, and miscellaneous conditions. *Sem Ped Surg* 2002; 11: 75-82.
10. Rintala RJ, Lindahl H. Is normal bowel function possible after repair of intermediate and high anorectal malformations? *J Pediatr Surg* 1995; 30: 491-494.
11. Langemeijer RATM, Molenaar JC. Continence after posterior sagittal anorectoplasty. *J Pediatr Surg* 1991; 26: 587-590.
12. Scharli AF. Anorectal incontinence: Diagnosis and treatment. *J Pediatr Surg* 1987; 22: 693-701.
13. Rintala RJ, Lindahl H. 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* 1999; 34: 334-337.
14. Kelly JH. Cine radiography in anorectal malformations. *J Pediatr Surg* 1969; 4: 538-546.
15. Varma KK. Long-term continence after surgery for anorectal malformations. *Ped Surg Int* 1991; 6: 32-35.
16. Hedlund H, Peña A, Rodriguez G, Maza J. Long-term anorectal function in imperforate anus treated by a posterior sagittal anorectoplasty: Manometric investigation. *J Pediatr Surg* 1992; 27: 906-909.
17. Husberg B, Lindahl H, Rintala R, Frenckner B. High and intermediate imperforate anus: Results after surgical correction with special respect to internal sphincter function. *J Pediatr Surg* 1992; 27: 185-189.
18. Holschneider AM, Ure BM, Pfrommer W, Meier-Ruge W. Innervation patterns of the rectal pouch and fistula in anorectal malformations: A preliminary report. *J Pediatr Surg* 1996; 31: 357-362.
19. Emblem R, Diseth T, Morkrid L. Anorectal anomalies: anorectal manometric function and anal endosonography in relation to functional outcome. *Pediatr Surg Int* 1997; 12: 516-519.
20. Yuan Z, Bai Y, Zhang Z, Ji S, Li Z, Wang W. Neural electrophysiological studies on the external anal sphincter in children with anorectal malformation. *J Pediatr Surg* 2000; 35: 1052-1057.