Colonic phasic motor activity is stronger in patients with repaired anorectal malformations than patients with severe colonic dismotility

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Abstract

In this study, colonic manometry studies of the patients with repaired anorectal malformations (ARM) were compared with those of patients with severe colonic dismotility due to chronic constipation (CC) and acute pseudo-obstruction (PSO). The patients with repaired ARM were accepted as group #1 (n=10). The patients with CC and acute PSO composed group #2 (n=10). Eight-channel water perfused catheter was inserted into the colon under sedation. Colonic activity was recorded in three phases including fasting, after meal and after bisacodyl installation. The results were assessed by Pearson χ^2 test, P<.05 was considered statistically significant. Mean age was 9.6 and 12.1 in groups #1 and #2, respectively. Ninety-five per cent of all patients had propagated contractions (PCs) and 20% and 40% of the patients in group #1 had PCs during fasting and after meal, respectively. These contractions were seen 30% and 70% of the patients in group #2, but no statistical difference was found between the groups. PCs after bisacodyl were observed 90% and 40% of the patients in groups #1 and #2, respectively, and this difference was statistically significant (P=.019). In this study, the prominent difference between the groups was found in response to intraluminal stimulation. This finding may indicate that the colon of the patients with ARM has more capacity to develop PCs by peripheral stimuli and more regular enteric nervous integrity.

Introduction

Colonic manometry has been increasingly used to manifest the motor activity of entire colon in children, especially after nineties.[1] The children with chronic constipation (CC) as an intractable colonic motility disorder constitute the most frequent pediatric population whose colonic activity is evaluated via colonic manometry. However, recently, new data has been obtained regarding other pediatric groups, such as repaired anorectal malformations (ARM), Hirschsprung's disease, spinal abnormality and cerebral palsy.[2-5]

Constipation and/or fecal incontinence are, now, a well-known problem of many patients with repaired ARM.[6] Seventy five percent of the patients with repaired ARM have voluntary bowel movements (VBM) and nearly half of them have fecal incontinence.[7] Although, there have been many manometric studies to evaluate anorectal region in these patients, information about colonic motility is still limited.

The aim of this study was to find out phasic motor activity of the colon in patients with ARM and to compare them with the results obtained from the children with severe colonic dismotility.

Materials and Methods

Patient population

Between February 2007 and September 2009, 10 patients with repaired ARM (group #1) and 10 patients with various colonic motility disorders (group #2) underwent colonic manometry. All studies were performed with the approval of the Ethical Committee of the Faculty of Medicine at Gazi University and after taking the consent of the patients' parents.

Group #1

Colonic manometry was done in two patients before the closure of the colostomy at an older age. Other patients had been already evaluated and accepted having impaired colonic motility. Four patients under bowel management program were examined before antegrade continent enema procedure (ACE). The other two patients were evaluated before sigmoid resection because of pseudo incontinence and the last two patients were investigated before bowel management program.

Group #2

Colonic manometry was performed in eight patients with severe CC unresponsive to previous treatments. Three of them had fecal incontinence in addition to CC. The other two patients underwent colonic manometry because of acute colonic pseudo-obstruction (PSO) owing to appendectomy and lymphoma.

Since the patients in both groups were involved in anorectal manometry studies in the past, they were not investigated again during colonic manometry.

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Colonic manometry

Colonic manometry was performed according to previously reported protocol.[1] The day before the study, bowel cleansing was done with oral sodium phosphate solution (Fleet Fosfo-Soda, Kozmed, Ankara, Türkey) or polyethylene glycol solution (Golytely, Braintree Laboratories Inc, Braintree, Mass, USA). A guide wire was inserted by using colonoscope and a flexible catheter was placed over the guide wire. The position of the catheter was verified by plain abdominal x-ray graphy. The colonic manometry catheters made by polyvinylchloride had a central lumen surrounded by 8 recording sites spaced 5, 10 to 15 cm apart. The catheters were water perfused and connected to polygraph and computer software (Medical Measurement Systems B.V., Version 8.3, the Netherlands). Colonic motility was evaluated two hours later or the day after the procedure following the patients' recovery from sedation or general anesthesia. Phasic contractile activity of the colon was recorded during 1 hour of fasting and 3 hours after the administration of a standard meal. Then, bisacodyl (0.2 mg/kg, max 10 mg) was infused through the central lumen, and

recording was continued for at least 40 min. Recorded activity was analyzed visually and interpreted by the same author. Propagated contractions (PCs) of the colon were defined as pressure waves migrating over 3 or more adjacent recording channels at a velocity of 0.2-12 cm s^{-1}. High-amplitude propagated contractions (HAPCs, >60 mmHg) and low-amplitude propagated contractions (LAPCs, <60 mmHg) as types of PCs were counted in all recording phases. PCs after the meal were thought as normal gastro colonic responses. When PCs were not seen at any phase of the study or at any segment of the colon, the motor activity was considered abnormal.

Analysis

For statistical analysis, Pearson χ^2 test (2-sided) was used, P<.05 was considered to be statistically significant. Statistical analysis was performed using SPSS 11.0 version.

Results

General properties of the groups are shown in Tables 1 and 2. The mean age during the study was 9.6 in group #1 and 12.1 in group # 2. Bowel preparation was achieved in all but three patients. Colonic manometry properties and findings are given in Table 3 for group #1 and in Table 4 for group # 2. The mean time of the studies was 5.3 and 6.7 hours for group #1 and group #2, respectively. Only for one patient in group #2 (Table 4), the study had to be stopped because of vomiting, severe abdominal pain and distention of the patient after bisacodyl installation.

The tip of the catheter on the x-ray was found at the cecum in 8, transverse colon in 2, hepatic flexura in 6, and splenic flexura in 4 of the patients. During the fasting period, PCs were found in 20% and 30% of the patients in group #1 and group #2, respectively. Gastrocolonic response after the meal was detected in 40% of the patients in group #1 and 70% of the patients in group #2. There was no statistical difference between the groups in terms of the PCs in fasting period (P=.606) and after the meal (P=.178). Ninety per cent of the patients in group #1 showed a response to intraluminal stimulation whereas only 40% of the patients in group #2 did so and the difference between the groups was found statistically significant (P=.019). Although, in general, the type of the recorded PCs was HAPCs, LAPCs were determined in 25% of all series. The appearance of HAPCs was different from the others in two patients with colostomy in group #1. These contractions looked like a motor migrating complex seen a motor activity of the small bowel in fact.

The propagated contractile activities belonging to group #1 were not seen in the rectum (1), rectosigmoid colon (2), rectosigmoid plus ascending (1) or descending colon

Table 1. General properties of group #1.

No	Sex	Age during the study	Type of ARM	Reason for colonic manometry
1	M	8	Rectovesical fistula	Before colostomy closure
2	F	18	Cloaca	Before colostomy closure
3	M	8	Perineal fistula	Before sigmoid resection
4	M	5	Perineal fistula	Before sigmoid resection
5	M	15	Rectourethral fistula	Before ACE procedure
6	M	7	Perineal fistula	Before bowel management program
7	F	9	Vestibular fistula	Before ACE procedure
8	F	12	Vestibular fistula	Before ACE procedure
9	M	10	Rectourethral fistula	Before ACE procedure
10	F	4	Perineal fistula	Before bowel management program

ARM, anorectal malformations; ACE, antegrade continent enema.

Table 2. General properties of group #2.

No	Sex	Age during the study	Reason for colonic manometry
1	M	12	Chronic constipation and fecal incontinence
2	F	13	Chronic constipation
3	M	7	Chronic constipation
4	M	11	Chronic constipation and before closure of sigmoid colostomy
5	F	19	Chronic constipation
6	F	15	Acute colonic PSO after appendectomy
7	M	6	Acute colonic PSO before diagnosis of mediastinal lymphoma
8	F	11	Chronic constipation and fecal incontinence
9	M	14	Chronic constipation and fecal incontinence
10	F	13	Chronic constipation

PSO, colonic pseudo-obstruction.

Table 3. Colonic manometry findings of the patients in group #1.

No	Duration of the study (hour)	Registered region of the colon	PCs during fasting	PCs after meal	Response to bisacodyl	Type and number of PCs HAPCs	LAPCs	Segmental absence of PCs
1	8	Distal to HF	+	-	+	23	-	
2	7	Distal to HF	+	-	+	30	-	
3	5	Total colon	-	+	+	12		RSC
4	5	Distal to SF	-	+	+		4	RSC
5	4	Distal to SF	-	+	+	1	-	
6	3.5	Distal to HF	-	-	+		1	DC, RSC
7	5	Total colon	-	-	+	5	-	
8	4.5	Total colon	-	-	+	2	-	
9	5.5	Total colon	-	+	-		1	AC, RSC
10	5.5	Distal to TC	-	-	+		1	Rectum

HF, hepatic flexura; TC, transverse colon; SF, splenic flexura; AC, ascending colon; DC, descending colon; RSC, rectosigmoid colon; HAPCs, high-amplitude propagated contractions; LAPCs, low-amplitude propagated contractions.

Table 4. Colonic manometry findings of the patients in group #2.

No	Duration of the study (hour)	Registered region of the colon	PCs during fasting	PCs after meal	Response to bisacodyl	Type and number of PCs HAPCs	LAPCs	Segmental absence of PCs
1	7	Total colon	-	+	+		22	Rectum
2	5.5	Distal to HF	-	+	-	3		-
3	18	Total colon	+	+	-	14		RSC
4	5	Proximal to SC	-	+	+	7		-
5	5.5	Distal to SF	-	+	-	2		RSC
6*	3	Distal to HF	-	+	-	2		-
7	8	Total colon	-	-	-	-	-	DC, RSC
8	5.5	Distal to HF	+	-	-	2		RSC
9	5	Distal to SF	+	+	+	13		RSC
10	5	Distal to TC	-	-	+	1		RSC

*Cancellation because of vomitus, severe abdominal pain and distention of the patient after bisacodyl. HF, hepatic flexura; TC, transverse colon; SF, splenic flexura; AC, ascending colon; DC, descending colon; RSC, rectosigmoid colon; HAPCs, high-amplitude propagated contractions; LAPCs, low-amplitude propagated contractions.

(1) of some patients (Table 3). The same pattern was also seen in group #2 (Table 4), including the rectum (1), rectosigmoid colon (5), rectosigmoid plus descending colon (1).

Patients' follow-up is briefly summarized in Table 5.

Table 5. Clinical follow-up of the groups.

	Treatment	Follow-up
Group #1		
1	-	Waiting for colostomy closure
2	Colostomy closure	VBM without soiling
3	Sigmoid resection	VBM without soiling
4	Stimulant laxatives	Waiting for sigmoid resection
5	ACE procedure	Without soiling for 24 hours
6	Bowel management program	Without soiling for 24 hours
7	ACE procedure	Without soiling for 24 hours
8	Bowel management program	Waiting for ACE procedure
9	Bowel management program	Waiting for ACE procedure
10	Bowel management program	Without soiling for 24 hours
Group #2		
1	Laxatives, then TNS	Unchanged
2	Sigmoid resection, then TNS	Unchanged
3	Sigmoid resection, then TNS	Unchanged
4	Sigmoid resection & Colostomy closure	Unchanged
5	Sigmoid resection	VBM with smearing
6	Erytromycin, neostigmin, then TNS	Gained VBM again
7	Chemo & Radiotherapy	Gained VBM again
8	Laxatives, then TNS	VBM without soiling
9	Laxatives, then TNS	VBM without soiling
10	Laxatives, then TNS	Unchanged

VBM, voluntary bowel movements; ACE, antegrade colonic enema, TNS, tibial nerve stimulation.

Discussion

In this study, the colonic motor activity of the patients with repaired ARM was compared with the patients who had severe colonic dismotility. Manometry was used for this purpose, thus, phasic contractile activity of the colon could be evaluated, directly. It is important because colonic motility effects the quality of life in patients with ARM after definitive surgery and there are not enough data about the contractile patterns of their colon in the literature. Heikenen *et al.* studied colonic manometry in thirteen patients with soiling after the repair of ARM for the first time (2). They found that all patients including both low and high types of ARM with or without constipation had HAPCs with an average of 80% propagation into the neorectum and proposed that excessive numbers of HAPCs could be a reason for fecal incontinence as well as sphincter dysfunction. In this series, although all patients in group #1 had PCs, HAPCs were found in six of them (60%) and the rest had LAPCs. In addition, PCs were capable of reaching to the rectosigmoid region in only half of the patients. The difference of HAPCs between the two series may result from the difference in the motility pattern of the colon. Since nearly half of the patients in mentioned study received loperamide for treatment, it could be considered that those patients had more hyper activated colon than the patients in this study. Furthermore, here, because of the dilatation, the PCs in that area might have been recorded as absent or LAPCs.

The response to intraluminal stimulation was the most important difference between the groups in this study. The patients with repaired ARM showed more phasic contractile activity than the others after bisacodyl (P=0.19). Therefore, the enteric nervous system of the colon in the patients with repaired ARM could be considered to have more regular integrity compared to acute or chronic functional motility problems of the colon. Van den Berg *et al.* used the colonic manometry as a predictor of cecostomy success in children with defecation disorders including idiopathic constipation, Hirschsprung's disease, ARM, cerebral palsy and spinal abnormality (5). When the findings of their study were inspected, it was seen that 60% of the patients with repaired ARM had positive bisacodyl response whereas this ratio was 35.7% in children with constipation. They reported that a colonic response with HAPCs after bisacodyl administration was found predictive of success and the absence of HAPCs in the entire colon was associated with unsuccessful outcome. Present study can be considered to be consistent with their findings. We observed that almost all patients with repaired ARM benefit from the treatment special for them; however, half of the patients with CC could not gain bowel habits despite intensive therapies (Table 5).

Two patients with colostomy in group #1 gave us a chance to evaluate unused distal colon. The frequency and the appearance of PCs in these patients were found different from the others. Probably, very frequent and intense HAPCs were recorded because of the fact that the narrowed lumen of the colon was excessively stimulated by water perfusion, and they seemed like a migrating complex.

To date, the reduced numbers of ganglia cells and/or glial cells, abnormal nerve fiber density in the circular muscle layer and a reduced number of interstitial cells of Cajal have been recognized in children with colonic motility disorders.[8-10] Colonic manometry has been used to discriminate these neuromuscular abnormalities of the colon; for instance, the weakness or absence of PCs have been thought to be due to muscular defects of the colon and uncoordinated HAPCs to be associated with neuropathic changes.[1,11] However, recently, it has been shown that there is a poor correlation between colonic manometry and histopathology, and specific manometric findings are unable to point to myopathic or neuropathic features.[12] Hence, the manometric results of the children in present series were difficult to be used for the explanation of underlying etiologic factors. In this study, colonic manometry was mainly used to decide segmental resection and colostomy closure even though it could not show all the motility patterns of the colon, such as tonic contractions.

Extrinsic and intrinsic abnormalities on the rectal wall in patients with ARM have been known for a long time.[13] However, there has not been any study showing a correlation between motility and histopathology yet. Recently, we reported that in children with repaired ARM and constipation, stasis is mostly seen in the rectosigmoid region regardless of VBM existence.[14] Despite the fact that there were absence of PCs and stasis in the rectosigmoid region, the proximal colon of the patients with repaired ARM was generally found to be more excitable compared to CC and acute PSO in this study. Small number of patients makes it difficult to propose whether phasic activity of the colon is related to ARM type.

The two patients in group #2 did not have any gastrointestinal problem before admittance to the hospital. These patients may cause heterogeneity in their group, however, our aim was to compare ARM patients not only with chronic functional motility disorders, but also acute colonic motility changes.

The patient with acute colonic PSO after appendectomy made us to consider Ogilvie's syndrome which results from the sudden unset of colonic dilatation without any mechanical obstruction.[15] Although the exact mechanism behind this idiopathic acute colonic PSO is not fully elucidated, the dissection around the cecum during appendectomy might have led to this condition. Intestinal PSO can be the first symptom of a tumor and it has been presumed to be due to inflammation of the myenteric plexus with destruction of the ganglion cells caused by antineuronal nuclear antibodies.[16] The first symptom of the case in this series was severe constipation in a short period, and mediastinal lymphoma revealed when the etiology of the constipation was searched. Resolution of the constipation after chemotherapy suggested a clinical manifestation like paraneoplastic syndrome.

In conclusion, the comparison of colonic manometry between the patients with repaired ARM and the patients with severe colonic dismotility in this study showed that colonic response to intraluminal stimulation was stronger in the ARM group than the others. This data supports that similar symptoms in the patients with colonic dismotility are resulted from different etiologic factors even if colonic manometry had limitations to find out underlying reasons. In future, larger and comparative series are necessary, thereby, therapeutic approach may be adjusted to these findings.

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