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Evaluation of anorectal function after transanal one-stage endorectal pull through operation in children with Hirschsprung's disease

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Abstract: Objective The short-term efficacy of the transanal one-stage endorectal pull through operation for Hirschsprung's disease is satisfactory. However the long-term outcome of anorectal function has not been fully understood. The aim of this study was to evaluate the stooling pattern, colonic motility and anal sphincter performance after transanal one-stage pull through operation in children with Hirschsprung's disease. **Methods** Fifty-eight children who underwent transanal one-stage pull through operation for Hirschsprung's disease were followed up. The mean follow-up duration was 15.8 months (range, 6-24 months). The stooling patterns of the patients were investigated by the informed questionnaire. Barium enema, defecography, total and segmental colonic transit time and the anorectal vector manometry were performed. Thirty-three healthy children were used as controls. **Results** Most of patients had normal stool consistency and frequency. Postoperative enterocolitis occurred in 3 patients, and constipation was found in five patients. Postoperative soiling was observed in 9 patients. None of the 58 patients had incontinence, cuff infection, anastomotic leak and mortality. The barium enema showed that the configuration of the colon recovered well in most of patients. Postopertive defecography showed the anorectal angle of all the patients was open, fixed and bigger than that of preoperation and the healthy controls ($P<0.01$). Postoperatively, the mean total gastrointestinal transit time(TGITT), the left colonic transit time (LCTT) and rectosigmoid colonic transit time (RSTT) in the 58 patients were significantly shorter than preoperatively ($P<0.01$) and were similar to those of the control group. The rectoanal inhibitory reflex was regained in 5 patients. The anal maximal pressure of the patients with constipation in resting and squeezing condition were significantly higher than those of the asymptomatic patients and controls ($P<0.05$). The vector volume (VV) and vector symmetric index (VSI) in patients with soiling were significantly lower than those in preoperation and the controls ($P<0.05$). The VSI in the patients with constipation was significantly higher compared with the controls ($P<0.05$). **Conclusions** The stooling function, colonic motility and anal sphincter performance manifest well in most of the patients after the transanal endorectal pull through operation for Hirschsprung's disease. Stooling disorders in few cases are probably related to decrease or disappearance of the sigmoid loops, dysfunction of the "neorectosigmoid", an open and fixed anorectal angle and achalasia of the internal anal sphincter.

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Key words: Transanal endorectal pull through; Hirschsprung's disease; Colonic motility; Anal sphincter; Child

经肛巨结肠根治术后患儿的肛肠功能评价

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[**摘 要**] **目的** 经肛门巨结肠一期根治术近期效果好，但术后患儿的肛肠功能目前尚不十分清楚。该文旨在评估患儿术后的排便模式，结肠和肛门括约肌功能。**方法** 对58例经肛门巨结肠根治术后半年以上儿童进行随访，随访时间为15.8月（6~24月）。并进行排便功能问卷调查、钡灌肠、结肠传输时间和肛门直肠测压检查。33例正常儿童作为对照。**结果** 大多数随访患儿排便正常，无任何临床症状。4例出现稀便便频，9例污便，5例便秘，3例小肠结肠炎。钡灌肠结果显示大多数随访患儿结肠形态恢复良好。全部病例术后直肠肛管角（度）较对照组显著开大，有症状组较无症状组显著开大。58例随访患儿的平均全胃肠、左半结肠和右半结肠传输时间较术前显著缩短，与对照组相比差异无显著性意义。直肠肛管反射5例阳性。便秘组的肛管最大静息压和最大收缩压明显高于无症状组和对照组。污便组向量容积和对称指数较对照组显著降低。便秘组对称指数显著高于对照组。**结论** 经肛门巨结肠根治术后大多数患儿排便功能、结肠功能和括约肌功能良好。少数病例排便功能障碍可能与

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术后乙状结肠曲减少或消失、“新直肠”储便功能代偿不全和拖出结肠致直肠肛管角开大、肛门括约肌痉挛失弛缓有关。

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[关 键 词] 经肛门巨结肠根治术；先天性巨结肠；结肠动力；肛门括约肌；儿童

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In 1998, Luis Dela Torre et al [1] firstly described the transanal endorectal one-stage pull through operation for Hirschsprung's disease. Since then, the procedure has become increasingly common[2,3]. Compared with the traditional laparotomy such as the Swenson, Duhamel and Soave procedure, the main advantage of the operation is that it is minimally invasive and eliminates the abdominal incision, so there is no abdominal scar and complications of traditional laparotomy postoperatively. The operating time and hospital delay are also cut short. However the long-term anorectal function of the patients after operation has not been fully understood. Although some research on the long-term outcome of anorectal function can be noted [4-10], the results varied widely. This study evaluated the stooling patterns, colonic motility and sphincter performance for the children who underwent the transanal one-stage pull through operation for Hirschsprung's disease by a follow-up investigation.

Materials and methods

Seventy-three children with Hirschsprung's disease underwent the transanal endorectal one-stage pull through operation between October, 1999 and July, 2002 at the 2nd Affiliated Hospital of China Medical University. A follow-up was performed on the children. Fifty-eight children were responded in a consecutive order and 15 were not, with a losing rate of 20%. All of them had an aganglionic segment confined to the rectosigmoid area confirmed by pathologic examination but none of them had a history of colostomy or open operation.

The series investigation of the stooling pattern was based on a personal or telephone interview documented with a questionnaire, including the following issues: age, sex, type of Hirschsprung's disease, occurrence of enterocolitis, times and characteristics of stools, constipation, continence, soiling, length of removed colon, and additional treatment of Hirschsprung-associated functional disorders.

The barium enema was performed on the 58 patients. The appearance of colon, sigmoid loops and enterocolitis were observed. Defecography was performed in 16 of the patients. The lateral films of the pelvis were taken at rest and during both squeeze and push, the anorectal angle was surveyed in different dynamic states [11]. The colonic transit time (CTT) was measured in the 58 patients using the simplified method of radio opaque markers (80% saturation method) described by Metcalf et al [8,12]. Anorectal manometry was performed on the 58 patients at rest in the left lateral decubitus position using the continuous pull-through method. The basal resting pressure (BRP) and maximal squeeze pressure (MSP) were measured in centimeters of water. The presence of the rectoanal relaxation reflex was examined by inflation of the rectal balloon with air (50 mL). The requirement for a positive rectoanal reflex was a fall of at least 25% from the basal pressure level after inflation of the balloon on three consecutive measurements.

Thirty-three healthy children without a history of gastrointestinal and endocrine and metabolic diseases who matched age and sex distribution to the patients were used as controls. The controls consisted of 21 boys and 12 girls (mean age = 5 years).

The student t test and Fisher exact test were used for statistical analysis. A value of $P < 0.05$ was considered statistically significant.

Results

Population

Of the 58 children, 39 were male and 19 were female, with the mean age of 24.7 months at operation. The mean follow-up duration was 15.8 months (range, 6-24 months).

Stool frequency

On average the stool times was 2.2 ± 2.0 per day in the 58 children. 1-2 times stool occurred in 54 cases per day, and 8-10 times in the 4 cases.

Stool consistency

Forty-six patients had normal stool consistency. Only 3 patients had muddy stools with foul smell. Nine patients had pasty stools or hard stools.

Complications

Postoperative enterocolitis occurred in 3 patients, and constipation was found in five patients. Postoperative soiling was observed in 9 patients. None of the 58 patients had incontinence, cuff infection, anastomotic leak and mortality.

According to the clinical investigation, overall 12

patients were symptomatic and the remaining 46 were asymptomatic.

Barium enema

The configuration of the colon recovered well and the typical appearances of Hirschsprung's disease such as dilated, shifting and spastic segments disappeared in all the 58 children. The colon was kept in a physiological trend. The sigmoid loops decreased or disappeared and were absolutely correlated to the length of removed colonic segment in operation ($r = 0.89$, $P < 0.05$.) Defecography showed the anorectal angle of all the patients was open, fixed and bigger ($123.3 \pm 15.1°$) than that of preoperation ($84.7 \pm 8.3°$) and the control group ($79.0 \pm 11.6°$) (both $P < 0.01$). The anorectal angle in the symptomatic group was bigger compared with that of the asymptomatic group ($135.6 \pm 15.9°$ vs $111.0 \pm 14.3°$, $P < 0.05$) (Figure 1).

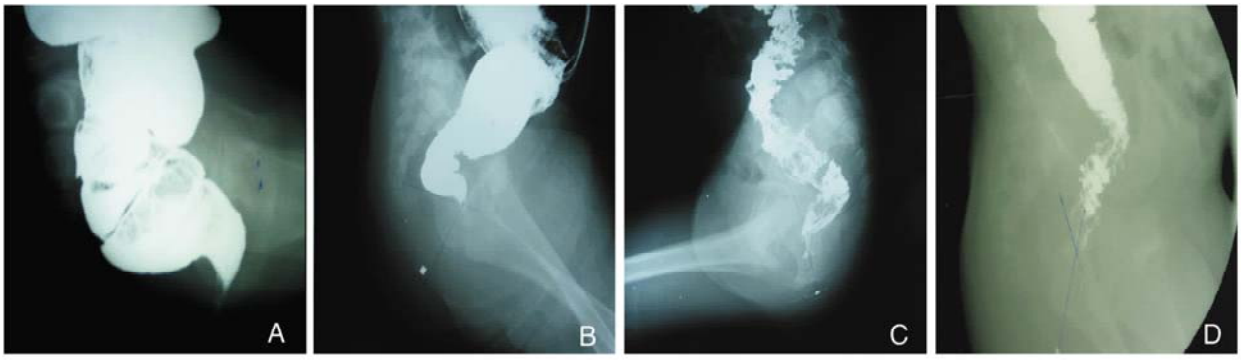

Figure 1 The barium enema appearances on the lateral side. **A**: Healthy control (male, 4 y). The colon was kept in a physiological trend. **B**: Preoperation (male, 5 y). The colon was kept in a physiological trend. However the spasm, shifting and dilated segment were noted. **C**: Postoperation (male, 4 y, no symptom). The sigmoid loops decreased and the anorectal angle was bigger than that of preoperation and the healthy control). **D**: Postoperation (male, 6 y, sufferring from frequent stools and soiling). The sigmoid loops disappeared and the anorectal angle was nearly linear.

Total and segmental colonic transit time

The mean total gastrointestinal transit time(TGITT), the left colonic transit time (LCTT) and rectosigmoid colonic transit time (RSTT) in the 58 patients were significantly shorter than preoperatively ($P < 0.01$) and were similar to those of the control group. The right colonic transit time (RCTT) in the 58 patients was not different from that in the preoperation. There were significant differences in the TGITT, LCTT and RSTT between the symptomatic and the control groups ($P < 0.01$). The TGITT, LCTT and RSTT of the asymptomatic group were not different from the control group (Table 1).

Vector manometry

The rectoanal inhibitory reflex (RIR) was regained in 5 patients (5/58, 8.6%). The anal maximal pressure of the constipated group in resting and squeezing condition were significantly higher than those of the asymptomatic and control groups ($P < 0.05$), while the anal maximal pressure of the soiling group was not different from the asymptomatic and control groups. The vector volume (VV) and vector symmetric index (VSI) were significantly lower in the soiling group than preoperatively and the control group ($P < 0.05$). The VSI in the constipated group was significantly higher compared with the control group ($P < 0.05$) but had no significant differences from that in preoperation (Table 2).

Table 1 The total and segmental colonic transit time ($h, \bar{x} \pm s$)

Group	n	TGITT	RCTT	LCTT	RSTT
Control	33	28.7 ± 7.7	7.5 ± 3.2	6.5 ± 3.8	13.4 ± 5.6
Preoperative	10	>168	9.1 ± 3.3	>60	>120
Postoperative	58	26.8 ± 8.2^{a}	7.6 ± 4.5	6.3 ± 4.1^{a}	11.8 ± 4.4^{a}
Symptomatic	12	$25.2 \pm 5.6^{a,b}$	7.7 ± 2.8	$6.0 \pm 4.2^{a,b}$	$9.8 \pm 4.0^{a,b}$
Asymptomatic	46	28.1 ± 10.1^{a}	7.4 ± 5.8	6.5 ± 3.4^{a}	12.2 ± 6.7^{a}

a Compared with the preoperative group, $P < 0.01$ (t test), b Compared with the control group, $P < 0.01$ (t test)

Table 2 Anorectal vector manometry findings ($\bar{x} \pm s$)

Group	n	Maximal pressure (mmHg)		VV	VSI
		Resting	Squeezing	(cm × cmHg²)	
Control	10	152 ± 33	190 ± 38	662 ± 311	0.70 ± 0.07
Preoperative	9	176 ± 38	236 ± 44	520 ± 254	0.75 ± 0.19
Postoperative	58	157 ± 47	200 ± 65	602 ± 312	0.70 ± 0.03
Constipation	5	167 ± 36[a]	211 ± 36[a]	638 ± 331	0.74 ± 0.02[a]
Soiling	9	151 ± 107	198 ± 102	381 ± 109[a]	0.69 ± 0.32[a]

a Compared with the control group, $P < 0.05$ (t test).

Discussion

This study was performed on a larger cohort of patients who underwent the transanal one-stage pull through operation at the Department of Pediatric Surgery, the 2nd Hospital, China Medical University. The results demonstrated the outcome of an unselected group of patients after surgery. An accurate follow-up was obtained in most patients by means of recent clinic visit. The results reveal that the transanal endorectal one-stage pull through operation is a safe and feasible procedure for Hirschsprung's disease.

This study found constipation, soiling and enterocolitis were common complications after surgery for Hirschsprung's disease. Compared with the reported data [4, 13-17], the morbidity of constipation was lower but the morbidity of soiling was higher in this study. Teitelbaum et al [15] described their experience with 78 infants undergoing primary pull through operation, which showed that the morbidity of constipation was 28%. Leeuwen et al [4] reported that the number was 22%. The variation may probably contribute to the proportion of age group, the race and the field or use of different criteria in different reports. There were 4.55% of the patients in this study experienced at least one episode of enterocolitis and required admission. The rate of enterocolitis varies widely from 54% to 1.4% [4,8,16,17]. The lower incidence of enterocolitis obtained in this study indicated that the transanal endorectal one-stage pull through operation may be a prospective technique for Hirschsprung's diseases. However a prospective and randomized study is needed to further prove it.

It has been reported that the outcome of colonic motility were not always satisfactory after surgery [8-10]. This study found the recovery of colonic motility located not only in the proximal colon but also in the distal colon after the transanal one stage pull through operation. The stooling disorder was noted in only a few cases.

The fact that the RSTT in the symptomatic children were significantly shortened revealed that the shortening of the colon is mainly located in the rectosigmoid area. This also confirmed that the stooling disorders might result from dysfunction of the "neorectosigmoid". On the other hand, the anorectal angle became open and fixed due to the colon pull through. The anorectal angle mainly reflected the function of the puborectalis that was considered as another important structure to control defecation [11,18]. But in this study, the pull through of the proximal colon made itself strain or even keep a little tension, and it was not retractable in the longitude. Moreover the pull through of the colon also made puborectalis ring full which opposed the puborectalis in the transverse section, so the anorectal angle became open and fixed. The puborectalis lost the function of stooling control and led to stooling disorders. Moreover, the pull through of the colon caused the colon and its mesentery pressed and strained, which could lead to ischemia of the colon. The colon ischemia increased not only the risk of anastomotic leakage, but also the risk of ischemic colitis that affected the colonic motility [19]. Controversy has existed concerning the absence or the presence of RIR during anorectal manometry after surgical procedures for Hirschsprung's disease. Leeuwen et al [4] reported that the RIR restored in 39%-91% of patients after surgery, but Heikkinen et al [5] considered that never or rarely did the RIR restore postoperatively. In this study, the RIR rate was 8.6%. This was similar to the previous reports which showed that the RIR presented in the majority of patients treated with the rectal myectomy but was absent in all of cases treated by the Swenson procedure [20, 21]. This suggested that the restoration of the RIR was originated from the conservation of muscular cuff in operation.

This study showed that the maximal resting pressure in the constipated children was higher than that of the control group suggesting that the constipated children had dysfunctions in the internal sphincter [22, 23]. For there was no internal sphincter at the bottom of the mobilized colon after surgery, the muscular cuff served as the internal sphincter and was often in dysfunction or achalasia without regulation of nerve. Furthermore, the enterocolitis from various uncertain causes might also lead to the dysfunction or achalasia. As a result, the maximal resting pressure grew higher, which destroyed the physiological asymmetry of pressure in anus. Therefore, dysfunction or achalasia of the anal sphincter and disappearance of the physiological asymmetry of

pressure in anus are probably the main reasons for constipation after surgery.

The VV and the VSI in the soiling children were significantly lower than those of the control and the asymptomatic groups, which showed that there existed anal sphincter injury in the soiling children, but the maximal resting pressure of the children was similar to the control and the asymptomatic groups. This suggested that the etiology of postoperative soiling was multifactorial. It might be caused by achalasia of the internal sphincter, which could lead to the improvement of the maximal anal pressure [22, 23] or injury from the posterior myotomy for the muscular cuff or from the dilated colon pull through the anal sphincter ring. It has been reported that many other sources are involved in the development of postoperative soiling [4,8].

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