



Patterns of postoperative enterocolitis in children with Hirschsprung's disease combined with hypoganglionosis

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Abstract

Objective: The relationship between postoperative enterocolitis (EC) and Hirschsprung's disease (HD) combined with hypoganglionosis (HYP) has not been thoroughly reported elsewhere. The aim of this study was to investigate the incidence of EC after operation in children with HD combined with HYP and to identify new strategies to prevent postoperative EC.

Methods: From 1998 to 2005, 97 children with HD underwent the modified Swenson's procedure in this institute. They were classified into 2 groups based on pathologic investigation as follows: group A contained 70 patients with HD and group B contained 27 patients with HD complicated with HYP. The mean follow-up time from the time of the operation was 3.4 years (range, 1.5-8 years). The incidence of postoperative EC and anorectal functions were evaluated and compared between these 2 groups.

Results: Eight cases (11.4%) in group A developed postoperative EC, whereas 11 (40.7%) in group B did so. The incidence of postoperative EC in group A was significantly lower than that in group B ($P < .005$). According to the Rintala scoring system, the percentage of patients with an excellent score in group A (85.7%) was significantly higher than that in group B ($P < .05$). The recurrence rate of constipation in group B was 14.8% (4/27), whereas it was 2.8% (2/70) in group A.

Conclusions: Postoperative EC is associated with retained proximal HYP, which suggests that HYP could be, at minimum, a predictive marker for this complication. Complete resection of HYP segment could potentially minimize the incidence of postoperative EC and alleviate the severity of EC.

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Enterocolitis (EC) is a significant complication of Hirschsprung's disease (HD) not only in preoperative periods but in postoperative periods. Enterocolitis can occur at any time from the neonatal period onward to adulthood and is independent of surgical procedure

performed. Despite many studies, the etiology is not completely understood. Recently, a study showed that surgical resection of intestinal neuronal dysplasia (IND) segment could decrease the occurrence of postoperative EC [1]. Pathologic diagnosis for the operative patients in our hospital showed that the incidence of hypoganglionosis (HYP) was higher than that of IND [2]. Schulten and coworkers [3] described that residual HYP increased the

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recurrent constipation rate and diarrhea rate. However, the relationship between the incidence of EC and HD combined with HYP has not been thoroughly investigated. Therefore, in this study, our goal was to evaluate the outcome of bowel function and the incidence of postoperative EC in children of HD treated by modified Swenson's procedure (heart-shaped anastomosis operation) [4] and to explore the relationship between postoperative EC and HD-associated HYP.

1. Materials and methods

1.1. Operation and diagnostic criteria

From 1998 to 2005, 70 children with HD (group A) and 27 with HD complicated with HYP (group B) underwent heart-shaped anastomosis procedure in our institute; the details of this procedure have been shown in our previous report [4]. The mean age at operation was 28 months old. All had constipation. All underwent preoperative evaluation including acetylcholinesterase (AChE) histochemical staining of rectal mucosal biopsy, anorectal manometry, and barium enema. Six patients in group A and 2 in group B had long segment aganglionosis; 56 in group A and 22 in group B had common segment; and 8 in group A and 3 in group B had short segment. The *aganglion segment* in the distalis of the middle rectum is defined as short segment HD, and the aganglion segment exceeding rectosigmoid is long segment and between them is common segment. All cases underwent modified Swenson's procedure with heart-shaped anastomosis, including the subtotal colectomy and left colectomy. The extent of bowel resection included both the narrow and dilated segment. Many of the patients had been previously treated incorrectly, so that the secondary lesion of intestine was severe, including a long dilated segment. Therefore, many patients having common segment, even short segment aganglionosis, underwent subtotal colectomy. The 2 resected bowel margins were submitted for pathologic examination after operation. The diagnosis of HD was based on that no neurons were detected the distal segment. And the diagnosis of HYP intestine was made in accordance to other published reports [2,5,6] as follows: area of nerve plexus reduced more

than half of normal, the number of neurons per unit length, and the diameter of the nuclei decreased, that is, less than 1.5 neurons per mm, but the cells were mature. Hirschsprung's disease associated with HYP was diagnosed based on that the distal segment was characterized by HD and the proximal segment with HYP.

1.2. Follow-up

The mean follow-up time was 3.4 years (range, 1.5-8 years). No one had postoperative anastomotic leak or stenosis.

A detailed questionnaire about the child's bowel function and EC was completed by the parents. The frequency of defecation, constipation, soiling, incontinence, and the incidence of diarrhea, abdominal distension, pyrexia, and vomiting were documented. Bowel function was investigated using Rintala scoring system [7], scores of 10 to 14 represented excellent, scores of 5 to 9 indicated fair, and scores of 0 to 4 was poor. Enterocolitis was diagnosed by clinical manifestations, such as diarrhea, abdominal distension, pyrexia, and vomiting, according to Carneiro et al [8]. Six children in group A and 3 in group B had preoperative EC.

1.3. Statistical analysis

Statistical analysis was performed using χ^2 test, and $P < .05$ was considered statistically significant.

2. Results

2.1. Preoperative diagnosis

All 97 patients presented with clinical manifestations of intractable constipation. In addition, some of them developed abdominal distension, intestinal obstruction, diarrhea, and vomiting before they went to the hospital. The results of clinical patterns, anorectal manometry, AChE staining, and barium enema before surgery are summarized in Table 1. In the barium enema study in group A, a narrow segment was noted in 62 (88.6%) patients, and barium stagnation for 24 hours was observed in 68 (97.1%). In group B, the same

Table 1 Results of preoperative objective routine examination of group A and B

Groups	Cases	RAIR absence, n (%)	Barium enema, n (%)		Positive AChE histochemical Staining, n (%)
			Narrow segment	Barium retention	
A	70	67 (95.7) *	62 (88.6)	68 (97.1)	68 (97.1)
B	27	22 (81.5)	23 (85.2)	26 (96.3)	24 (88.9)
Overall	97	89 (91.8)	85 (87.6)	94 (96.9)	92 (94.8)

RAIR indicates rectoanal inhibition reflex.

* $P < .05$.

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