Factors Conducive to Catch-Up Growth in Postoperative Jejunoileal Atresia Patients as Prognostic Markers of Outcome

Factors conductive to catch-up growth in postoperatve jejunoileal atresia patients as prognostic markers of outcome

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INTRODUCTION

Jejunoileal atresia (JIA) is a major cause of neonatal intestinal obstruction and requires resection of part of the small intestine during the neonatal period [1,2]. Morbidity and mortality associated with the management of neonates with JIA has improved [3,4] because of advancements in neonatal intensive care technology, refinement of operative techniques, use of total parenteral nutrition, and innovations in anesthesia. Despite these favorable factors, the postoperative course of JIA patients can often be impaired by poor weight gain that can affect quality of life. Thus, close follow-up after surgery has been advocated for ensuring stable growth during the immediate postoperative period [5]. Accelerated growth to regain normal size, i.e., catch-up growth (CUG) usually occurs as a distinct clinical entity within 2 years of surgery for JIA according to an unknown mechanism.

There are studies in the literature addressing the implications of surgery for JIA,

focusing on perioperative and short-term sequelae of surgery in neonates and young infants [6-8], but only a few studies about long-term outcome of JIA surgery [9,10]. The aim of this study was to assess postoperative growth in our JIA patients and identify factors associated with CUG.

MATERIALS AND METHODS

We reviewed the medical records of patients who underwent surgery for JIA at our institution between 1980 and 2013 (n=93). Patients with duodenal atresia and concomitant congenital anomalies (i.e., cardiac or central nerve system) were excluded. Of these, 42 patients with intact medical records were the subjects for this study. Patients were classified into 3 groups according to weight in comparison with standard Japanese growth and weight charts for children matched for age and sex [11]; group M+: more than mean at 12 months after surgery (n=13), group M-CUG+: less than mean at 12 months after surgery but more than mean at 24 months after surgery because

of CUG (n=11), and group M-CUG-: less than mean at 24 months after surgery because there was no CUG (n=18) (Figure1).

Figure 1

Parameters assessed were gestational age, birth weight, postoperative weight (on completion of surgery and at 1, 2, 3, 6, 12, and 24 months after surgery), site of JIA, length of residual small intestine measured intraoperatively during initial surgery for JIA, ratio of the length of residual small intestine to predicted length of small intestine for children of matched gestational age and sex (RP ratio), duration of parenteral nutrition, and incidence of postoperative complications such as cholestasis and sepsis. PLSI the predicted length of small intestine was obtained from existing data collected from normal neonates matched [12]. A weight standard deviation (WSD) score reflecting the extent of variance of subject weight from standard weight for age and sex matched normal infants was calculated for each subject and mean scores were compared between groups.

Data were expressed as mean ± standard deviation. Statistical analysis was performed using SPSS version 14.0 (SPSS, Chicago, IL), using the one-way ANOVA

test and the Tukey-Kramer test for post-hoc analysis and the Chi-squared test for analyzing categorical data. Differences were considered statistically different if $p < .05$. This study was approved by the Juntendo University School of Medicine Institutional Review Board and complies with the Helsinki Declaration of 1975 (revised 1983).

RESULTS

All surgery was successful without any complications. Demographic data are summarized in Tables 1 and 2. Results for the 3 groups (M+, M-CUG+, M-CUG-) were similar for gestational age (37.6±1.6 weeks vs. 38.2±1.5 weeks vs. 36.7±2.4 weeks; p=0.12), birth weight (2.9±0.3 kg vs. 2.7±0.4 kg vs. 2.6±0.5 kg; p=0.12), and incidence of jejunal atresia 5/13 (38%) vs. 2/11 (18%) vs. 7/18 (38%), respectively. Ileocecal valves were included in ileocecal resection for JIA of the distal ileum in 2 cases and both were in group M-CUG-. Figure 2 shows changes in postoperative weight.

Table 1

Table 2

Figure 2

There were no significant differences in the length of residual small intestine

between the 3 groups (126.0±34.5 cm vs. 121.6±44.7 cm vs. 109.6±41.5 cm; p=0.51, respectively). RP ratios were significantly higher in M+ (0.85±0.15) and M-CUG+ (0.84±0.18) compared with M-CUG- (0.69±18; p=0.02) (Figure. 3). However, there were significant differences in WSD scores at birth (0.92 ± 1.24 SD vs. -0.16 ± 0.91 SD vs. 0.39 ± 0.77 SD; p= 0.03, respectively). In particular, the WSD score at birth for M-CUG+ was significantly lower than for M+ (p=0.03). There was no significant difference in duration of parenteral nutrition between the 3 groups (29.7±23.7 days vs. 86.3±131.9 days vs. 62.2±157.0 days; p=0.54). The longest duration of parenteral nutrition was 690 days in one of the patients with no ileocecal valve. All patients had been weaned off parenteral nutrition by 2 years of age. Cholestasis associated with parenteral nutrition (serum level of conjugated bilirubin>2mg/dL) occurred in 3 patients (7%) and subsided without intervention.

Figure 3

DISCUSSION

To the best of our knowledge, our study is the first to identify a correlation

between RP ratio and CUG in postoperative JIA patients. While there are studies evaluating postoperative growth in patients with short bowel syndrome due to necrotizing enterocolitis, gastroschisis, mid gut volvulus, and other gastrointestinal conditions, there are few studies about JIA [13,14] and postoperative growth is not mentioned specifically because of a general assumption that normal growth will ensue. However, when we assessed our JIA patients, we found 43% weighed less than the mean for their age even after 2 years which was dismissed by parents and health care professionals as a consequence of major surgery as a neonate and not treated.

WSD scores were significantly lower in M-CUG+ compared with M+ and were significantly higher in M-CUG- compared with M-CUG+, indicating that patients with intrauterine growth retardation have a tendency to short-term growth retardation initially, but with proper care, later growth will be adequate and low birth weight is not detrimental to growth in the long-term.

Short residual small intestine and lack of an ileocecal valve have been well recognized as important factors affecting growth. Calisti et al suggested that length of

residual small intestine is correlated with requirement for parenteral nutrition and recommended primary end-to-end anastomosis to maximize the length of residual small intestine with plication of dilated proximal bowel if there was a caliber change not suitable for anastomosis [14]. There are also studies that evaluated the ratio of residual to predicted length of the entire small intestine in short bowel syndrome patients. For example, Spencer et al concluded that an intact ileocecal valve and residual small intestine more than 10% of the predicted length of the entire small intestine are predictors for weaning from parenteral nutrition [15].

Wales et al showed in a cohort study that presence of an ileostomy and length of residual small intestine less than 50% of the predicted length of the entire small intestine were associated with the development of short bowel syndrome [13]. According to their results, the ratio of residual to predicted lengths of small intestine (our RP ratio) seems to be of value for predicting future growth of patients after small bowel resection. However, these studies did not take into consideration the part of bowel that was resected or the gestational age of patients, both of which can greatly

affect bowel function so their scope is different to study, but we agree with their findings and avoid ileostomy whenever possible.

Intestinal length rapidly increases during late gestation, with a documented 1.5 fold increase in length occurring between the 30th and 40th weeks of gestation [16]. This acceleration in intestinal growth offer infants adequate absorptive capacity to meet postnatal nutritional demands. Although preterm infants are born without undergoing this accelerated lengthening, intestinal growth continues during early postnatal life and preterm infants have a greater potential for increasing length and mucosal surface area of the small intestine after birth than term infants. In fact, it has been suggested that preterm infants have more intensive intestinal function than term infants when the same length of small intestine are compared [17]. With this as background, it is of interest that the RP ratio was correlated to CUG while he actual length of residual small intestine was not relevant to CUG. While the predicted length of small intestine is dependent on gestational age, the length of residual small intestine is dependent on the extent of excision performed and may or may not be adjustable depending on the

location of pathology. Thus, the RP ratio gives a realistic quantification of the length of bowel available for CUG. Our recommendation of an RP ratio of at least 70% is based on the RP ratio in M-CUG- being less than 70% with significant differences in outcome in relation to M+ and M-CUG+. This arbitrary cut-off does not mean if the RP ratio is less than 70% there will be no CUG; it indicates there is poorer potential for CUG because there is less residual bowel with a lower volume of mucosa and capacity for adaptation and compensation. Conversely, a higher RP ratio would suggest there is a larger volume of mucosa available for adaptation and compensation which contributes to CUG in postoperative JIA patients.

We are conscious of 2 limitations that may affect our findings, namely, the effect of losing an ileocecal valve and only outpatient height and weight records available for assessing growth. Ideally we would like to review more patients without ileocecal valves because in this series there were only 2, and both were in M-CUG-, and we would like to include a review of postoperative nutrition in future follow-up studies.

CONCLUSION

Despite improvements in surgical management of JIA, outcome is marred by poor growth because of inadequate CUG. In this study, we found higher RP ratios were associated with CUG, while lower RP ratios were not and actual length of residual small intestine was of no prognostic value. RP ratios over 70% are recommended and with this background knowledge, surgeons can plan surgery appropriately to ensure patients achieve this level and thus have the best potential for CUG or patients with low RP ratios can be identified for more intensive follow-up with dietary and lifestyle counselling to enhance their potential for CUG.

FIGURE LEGENDS

Table 1: Demographics of our JIA patients

JIA = jejunoileal atresia

Table 2: Comparison of the 3 groups

WSD score = a weight standard deviation score

RP ratio = the ratio of residual to predicted lengths of small intestine

Figure 1: Classification of groups

mo. = months after surgery

42 patients who underwent surgery for jejunoileal atresia were divided into three groups according to weight compared with standard weights for matched controls; group M+: > mean at 12 months after surgery (n=13), group M-CUG+: < mean at 12 months surgery but >mean at 24 months because of CUG (n=11), and group M-CUG-: < mean at 24 months surgery because there was no CUG (n=18)

Figure 2: Changes in postoperative weight

SD = standard deviation, WSD score = a weight standard deviation score

Group M+ showed early CUG 3 months after surgery and group M-CUG+ showed late CUG 12 months after surgery, while group M-CUG- had no CUG.

Figure 3: Significance of RP ratios

RP ratio = the ratio of residual to predicted lengths of small intestine

RP ratios in M+ (0.85 ± 0.15) and M-CUG+ (0.84 ± 0.18) were significantly higher than M-CUG- (0.69 ± 18; p=0.02)

REFERENCE

1 Best KE, Tennant PW, Addor MC et al. Epidemiology of small intestinal atresia in Europe: a register-based study. Arch Dis Child Fetal Neonatal Ed 2012; 97: F353-358

2 Hemming V, Rankin J. Small intestinal atresia in a defined population: occurrence, prenatal diagnosis and survival. Prenat Diagn 2007; 27: 1205-1211

3 Stollman TH, de Blaauw I, Wijnen MH et al. Decreased mortality but increased morbidity in neonates with jejunoileal atresia; a study of 114 cases over a 34-year period. J Pediatr Surg 2009; 44: 217-221

4 Kumaran N, Shankar KR, Lloyd DA et al. Trends in the management and outcome of jejuno-ileal atresia. Eur J Pediatr Surg 2002; 12: 163-167

5 Olieman JF, Poley MJ, Gischler SJ et al. Interdisciplinary management of infantile short bowel syndrome: resource consumption, growth, and nutrition. J Pediatr Surg 2010; 45: 490-498

6 Piper HG, Alesbury J, Waterford SD et al. Intestinal atresias: factors affecting clinical outcomes. J Pediatr Surg 2008; 43: 1244-8

7 Prasad TR, Bajpai M Intestinal atresia. Indian J Pediatr 2000; 67: 671-8

8 Sato S, Nishijima E, Muraji T et al. Jejunoileal atresia: a 27-year experience. J Pediatr Surg 2008; 43: 1244-8

9 Dalla Vecchia LK, Grosfeld JL, West KW et al. Intestinal atresia and stenosis: a 25-year experience with 277 cases. Arch Surg 1998; 133: 490-496; discussion 496-497

10 Walker K, Badawi N, Hamid CH et al. A population-based study of the outcome after small bowel atresia/stenosis in New South Wales and the Australian Capital Territory, Australia, 1992-2003. J Pediatr Surg 2008; 43: 484-488

11 Ministry of Education, Culture, Sports, Science and Technology in Japan (2010) Mean Value and Standard Deviation of Height, Weight, and Seated Height. Statistical Survey of School Health 2010 (In Japanese)

12 Struijs MC, Diamond IR, de Silva N et al. Establishing norms for intestinal length in children. J Pediatr Surg 2009; 44: 933-938

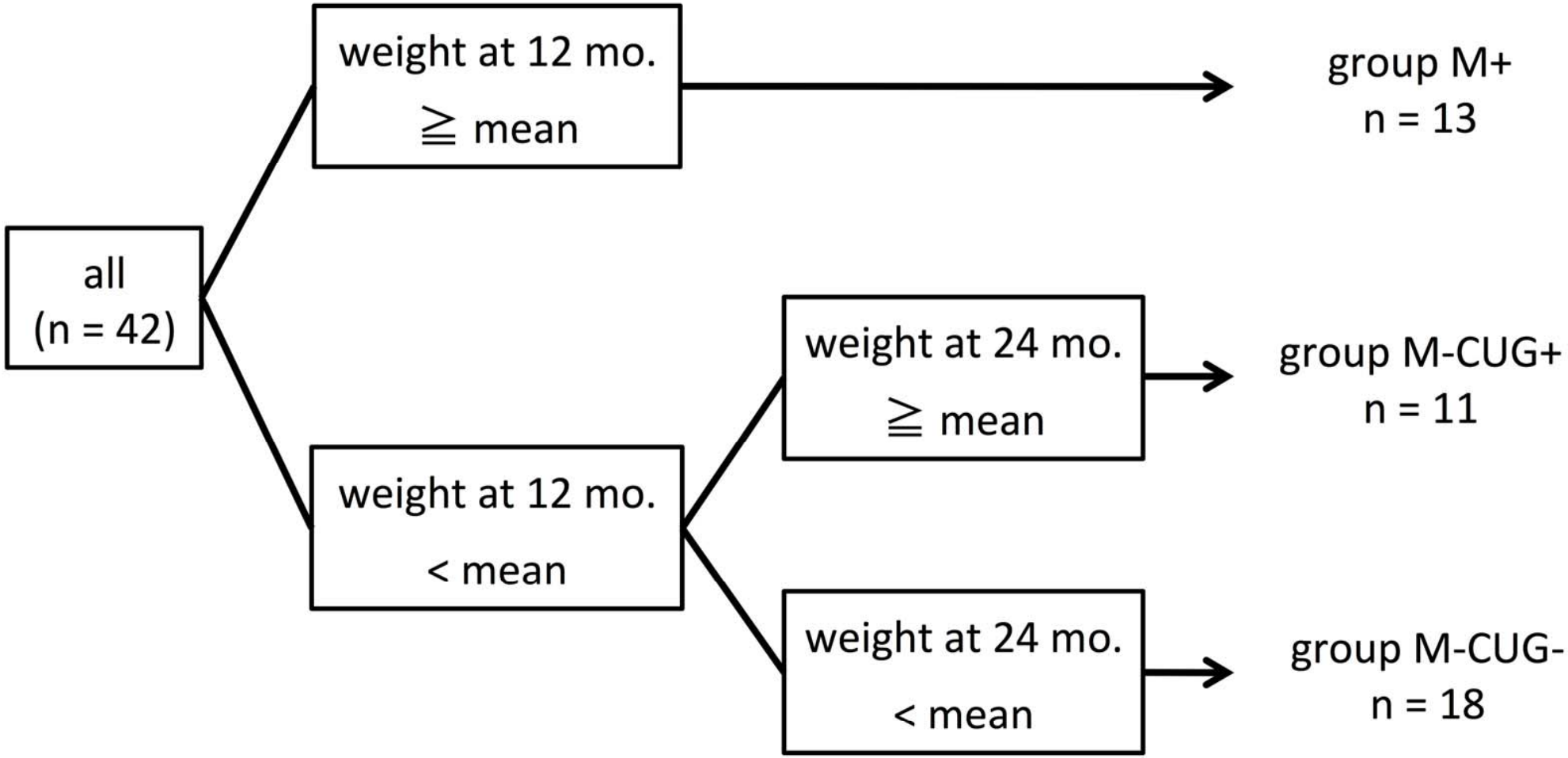
13 Wales PW, de Silva N, Kim JH et al. Neonatal short bowel syndrome: a cohort study. J Pediatr Surg 2005; 40: 755-762

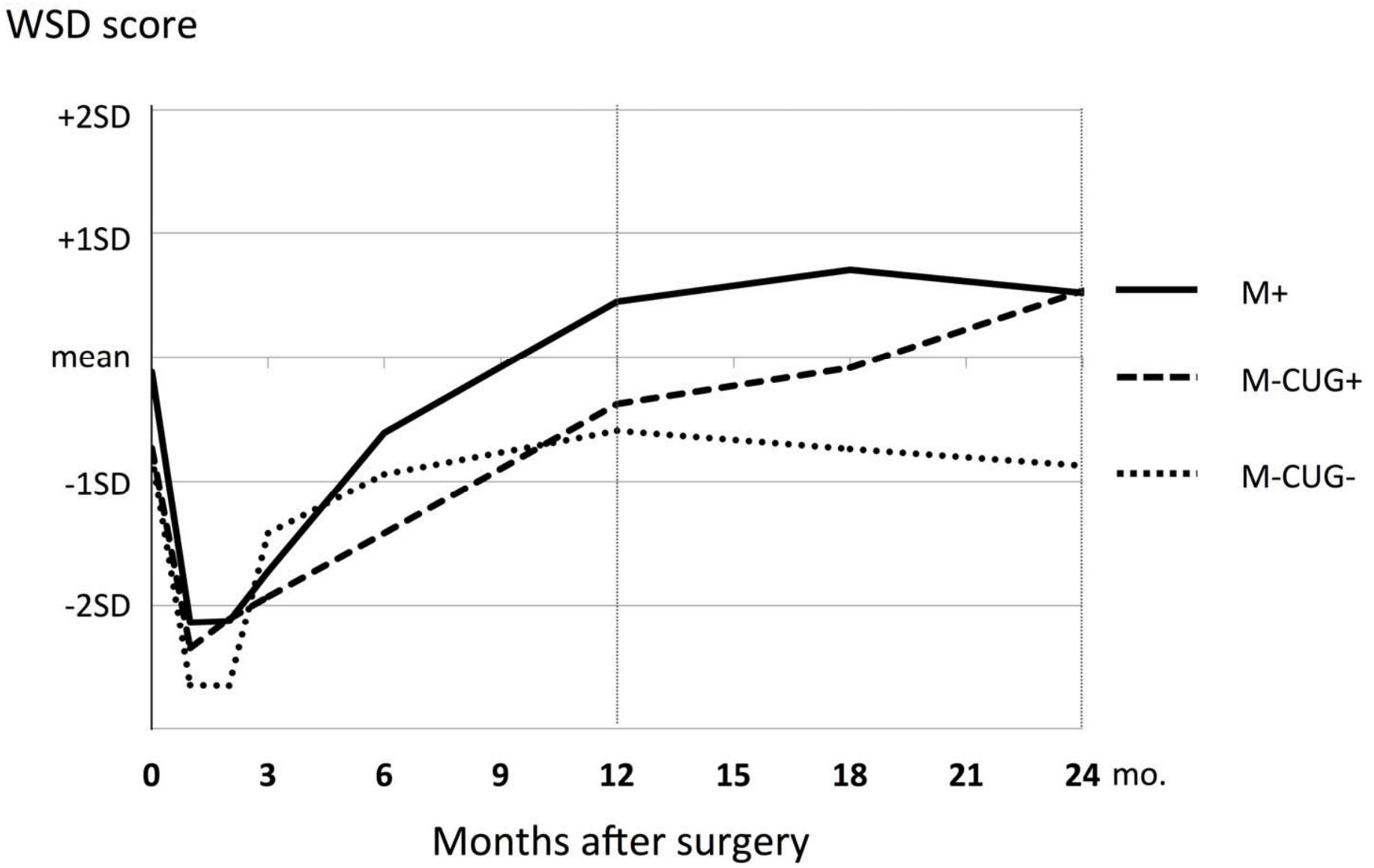
14 Calisti A, Olivieri C, Coletta R et al. Jejunoileal Atresia: Factors Affecting the Outcome and Long-term Sequelae. J Clin Neonatol 2012; 1: 38-41

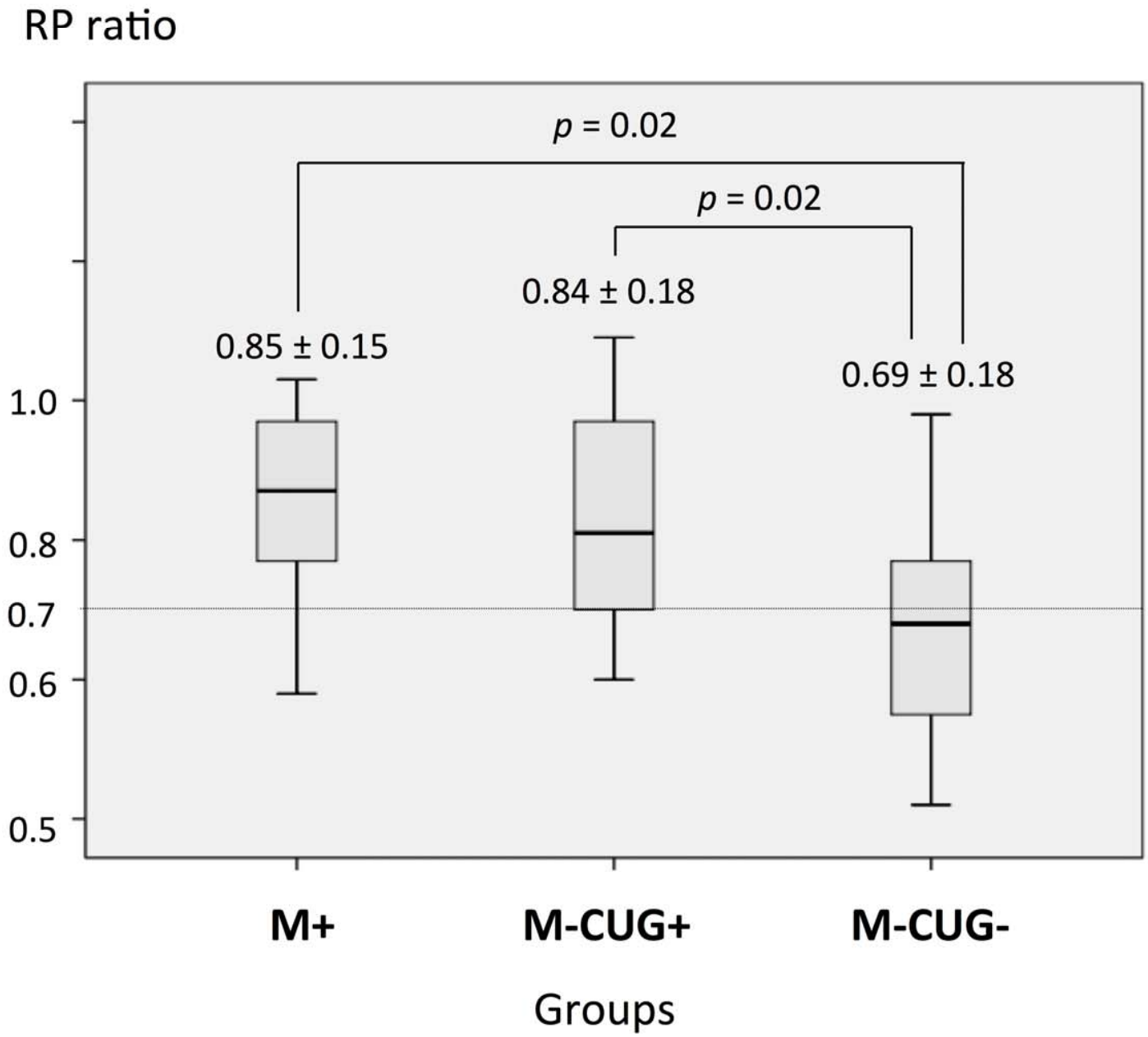
15 Spencer AU, Neaga A, West B et al. Pediatric short bowel syndrome: redefining predictors of success. Ann Surg 2005; 242: 403-409; discussion 409-412

16 Weaver LT, Austin S, Cole TJ. Small intestinal length: a factor essential for gut adaptation. Gut 1991; 32: 1321-1323

17 Touloukian RJ, Smith GJ. Normal intestinal length in preterm infants. J Pediatr Surg 1983; 18: 720-723


all
(n = 42)
weight at 12 mo.
≧ mean
group M+
n = 13
weight at 12 mo.
< mean
weight at 24 mo.
≧ mean
group M-CUG+
n = 11
weight at 24 mo.
< mean
group M-CUG-
n = 18


WSD score
+2SD
+1SD
mean
-1SD
-2SD
M+
M-CUG+
M-CUG-
0
3
6
9
12
15
18
21
24 mo.
Months after surgery


RP ratio
p = 0.02
p = 0.02
0.85 ± 0.15
0.84 ± 0.18
0.69 ± 0.18
1.0
0.8
0.7
0.6
0.5
M+
M-CUG+
M-CUG-
Groups

	Number of patients, (%) (n = 42)
Sex	
Male	22 (52)
Type of atresia	
Type Ⅰ	6 (14)
Type Ⅱ	12 (29)
Type Ⅲa	15 (36)
Type Ⅲb	5 (12)
Type Ⅳ	4 (9)
Site of atresia	
jejunum	14 (33)
ileum	28 (66)
Prenatal diagnosis	
confirmed	26 (62)

	Group M+ (n = 13)	Group M-CUG+ (n = 11)	Group M-CUG- (n = 18)	p value
Gestational age (weeks)	37.6 (±1.6)	38.2 (±1.5)	36.7 (±2.4)	0.12
Birth weight				
actual birth weight (kg)	2.9 (±0.3)	2.7 (±0.4)	2.6 (±0.5)	0.12
WSD score (SD)	0.92 (±1.24)	-0.16 (±0.91)	0.39 (±0.77)	* 0.03
Site of atresia				
jejunal atresia (n), (%)	5, (13)	2, (18)	7, (38)	0.9
Length of residual small intestine				
actual length (cm)	126.0 (±34.5)	121.6 (±44.7)	109.6 (±41.5)	0.51
RP ratio	0.85 (±0.15)	0.84 (±0.18)	0.69 (±0.18)	* 0.02
Duration of parenteral nutrition (days)	29.7(±23.7)	86.3 (±131.9)	62.2 (±157.0)	0.54