Gastro-Intestinal Conditions

Management of recurrent ileocolic intussusception[☆],[☆☆]

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A B S T R A C T

Purpose: The aims of this study were to evaluate the need for surgical intervention for patients with recurrent ileocolic intussusception (RICI), especially for multiple recurrences, and to investigate whether early and late recurrence patterns were associated with surgery.
Methods: Patients with ileocolic intussusception (ICI) during the years 2007–2019 were included. Demographic data, recurrences, and outcomes were analyzed. Early RICI was defined as recurrence within 48 h.
Results: Overall, 604 episodes of ICI were confirmed in 491 patients. The recurrence rate was 13.8%, with 113 episodes in 68 patients. There were no statistically significant differences in age, reduction success rate, operation, or pathological lead points (PLPs) between the recurrence and non-recurrence groups. There was no significant association between the number of recurrences and the presence of a PLP or between the number of recurrences and whether the recurrences were early or late. The presence of PLPs was not significantly associated with age or recurrence, but the reduction success rate was significantly lower ($P < 0.001$).
Conclusions: Each recurrence should be managed as a first episode, regardless of early or late recurrence. Operative reduction should be considered when nonoperative reduction fails, a PLP is suspected, or there are signs of peritonitis.
Type of Study: Treatment Study.
Level of Evidence: Level III.

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Ileocolic intussusception (ICI) is the most common cause of intestinal obstruction in children aged 3 months to 5 years [1]. In the United States, there are 56 pediatric cases of ICI per 100,000 children per year [2]. Around 90% of the cases are idiopathic, with no other pathological factors apart from lymphoid follicle hyperplasia at the end of the ileum. However, in some cases, invagination is secondary to an identifiable cause, such as Meckel's diverticulum, intestinal polyps, or benign or malignant intestinal tumors [3,4].

Nonoperative reduction of ICI by air or saline enema has a high success rate and few complications [5,6]. For this reason, nonoperative reduction is typically the first-line therapy for an initial episode of ICI. Surgical treatment is performed when nonsurgical treatment has failed or contraindicated, for example because of signs of perforation

(e.g., peritonitis, or pneumoperitoneum on a plain abdominal x-ray) or hemodynamic instability despite adequate resuscitation.

The most common complication after successful nonoperative reduction is recurrence, reported to occur in up to 20% of cases [7,8]. However, the optimum treatment for recurrent ICI (RICI) remains unclear. Some authors have recommended surgical intervention after more than one or two episodes of ICI [9,10], whereas, others have recommended that each recurrence is treated as though it was the first episode [11,12]. In cases of multiple recurrences, the surgeon is concerned about the possibility there may be a pathologic lead point (PLP) or bowel damage, and as well as the possibility of further recurrence. As ICI can be treated safely and effectively with the air or saline enema, the surgeon became more concerned with how many times to repeat nonoperative reduction in what is presumed to be idiopathic intussusception.

Most previous studies of RICI have focused on the clinical and ultrasound parameters that may predispose patients to recurrence or bowel damage [8,13,14]. Few studies have considered the surgical criteria for RICI or have observed the progression in patients with multiple recurrences [15]. In addition, few authors have distinguished between early and late recurrences. The aims of the present study were to evaluate

Table 1
Recurrence rate by age.

Age at the 1st episode	No. of cases	No. of recurrence cases (%)	P value[*]
0-1 y	129	13 (10.1)	0.46
1-2 y	173	28 (16.2)	0.32
2-3 y	118	17 (14.4)	0.57
3-4 y	48	6 (12.2)	0.34
4-5 y	16	3 (18.6)	0.16
>5 y	7	1 (14.3)	0.57
Total	491	68 (13.8)	

[*] Comparing the recurrence rate of each age group with the overall recurrence rate.

the need for surgical intervention for patients with RICI, especially for multiple recurrences, and to investigate whether early and late recurrence patterns were associated with surgery.

1. Methods

This retrospective study reviewed the data for children aged 0–15 years who were treated at Ulsan University Hospital with a diagnosis code for intussusception between January 2007 and January 2019. The hospital has a tertiary pediatric department that is the only referral center in Ulsan Metropolitan City. The Institutional Review Board of Ulsan University Hospital approved this study (2019-08-042).

Only patients with intussusception confirmed by ultrasound or Computed Tomography (CT) were evaluated. Four hundred eighty patients were diagnosed by ultrasound and 11 were diagnosed as initial CT without ultrasound. Thirty-four patients underwent CT to find a PLP after ultrasound and five patients underwent a Meckel's scan. Only those with ICI were included in the analysis, patients with ileo-ileal, jejuno-jejunal, or colo-colonic intussusception were excluded. The data collected for each patient included demographic data, clinical data, the total number of intussusception episodes within the study period, the outcomes of air enema and any operative procedures, and imaging and histopathologic findings.

Patients with no symptoms of peritonitis and who were hemodynamically stable were treated using the standard nonsurgical reduction technique. The method of nonoperative reduction in Ulsan University Hospital was air enema, which was performed by a radiologist under fluoroscopic or ultrasound guidance. Indications for surgery included failed reduction attempts, peritonitis on physical examination, signs of sepsis, evidence of perforation on imaging or during attempted enema, or significant concern that there may be a PLP. Surgery was performed by two pediatric surgeons. In 2007-2018, a senior surgeon performed open surgery, and in 2017-2019, a junior surgeon performed surgery using a laparoscopic approach.

Successful reduction was defined by records in the patient's notes that the passage of air into the small bowel loops was achieved and the intussusception seen on the initial images disappeared. Early RICI

was defined as recurrence within 48 h after successful ICI reduction and late RICI as recurrence after 48 h. A PLP was defined by the finding of a pathologic abnormality in the resected specimen that served as the lead point for the intussusception. A finding of ischemia and/or necrosis without a co-existing lead point was not considered to be a PLP.

The statistical analysis was performed using SPSS version 23.0. The data are summarized as count and percentage for categorical data and median and interquartile range for continuous data. Continuous parametric data were checked for normality and homogeneity of variance and analyzed using Student's t tests; noncontinuous data were analyzed using chi-square or Fisher's exact tests. A p value < 0.05 was considered statistically significant.

2. Results

During the 12-year study period, 604 episodes of ICI were confirmed in 491 patients. Of these 491 patients, 68 patients experienced RICI, with a total of 113 recurrent episodes. Thus, the overall recurrence rate was 13.8%. The age of the 491 patients at the first intussusception ranged from 2 months to 7 years (median, 21.3 months). The recurrence rates for the various age groups are shown in Table 1. There was no significant association between the recurrence rate and age.

The 68 RICI patients were considered to be the recurrence group and the remaining 423 patients as the non-recurrence group. There were no statistically significant differences between the recurrence and non-recurrence groups in age, reduction success rate, proportion who underwent surgery, or the presence of a PLP (Table 2). The 113 recurrent episodes included 53 early recurrence episodes and 60 late recurrence episodes. There were no significant differences between early and late recurrence episodes in reduction success rate, proportion of patients who underwent surgery, or the presence of a PLP. Among the 68 patients with RICI, 44 experienced a single recurrence, 15 experienced two recurrences, 3 experienced three recurrences, 2 experienced four recurrences, 3 experienced five recurrences, and 1 experienced seven recurrences (Table 3). There was no significant association between the number of recurrences experienced by a patient and the presence of a PLP (p = 1.0) or between the number of recurrences and the rates of early and late recurrence (p = 0.40). Of the patients who experienced two or more recurrences, seven experienced only early recurrences, seven only late recurrences, and 11 both early and late recurrences.

Overall, 20 patients required surgery, 18 after the failure of reduction, one because of a perforation during the air enema, and one because a PLP was suspected. Of these, 13 underwent intraoperative reduction and the other seven underwent bowel resection. The median age of the patients who underwent surgery was 21 months (range, 2 months to 6 years). A 6-year-old child was diagnosed with Henoch–Schönlein purpura and underwent a bowel resection because of bowel ischemia without a PLP. Three patients developed a recurrence after surgical

Table 2
Statistical analysis of the clinical features and outcome in 491 cases of ICI and 113 recurrent episodes.

	Non-Recurrence group (n=423)	Recurrence group (n=68)	P value	Early recurrence (episodes=53)	Late recurrence (episodes =60)	P value
1st episode age (months, range)	21.1 (2-85)	22.3 (2-60)	0.52			
1st episode age			0.16			0.45
<1 y (n)	115	13		7	12	
≥1 y (n)	308	55		46	48	
Sex (M:F)	267:156	46:22	0.47	31:22	44:16	0.09
Reduction success rate (%)[*]	96.5	92.6	0.18	96.2	98.3	0.6
Success (n)	408	63		51	59	
Fail (n)	15	5		2	1	
Operation (n)	15	5	0.18	3	1	0.34
Bowel resection (n)	5	2	0.25	1	1	1.0
PLP (n)	3	2	0.14	1	1	1.0

ICI; ileocolic intussusception, PLP; pathologic lead point.

[*] air enema reduction.

reduction; all were late recurrences and were successfully reduced by air enema. No patients experienced a recurrence of intussusception after bowel resection. There was no mortality and only one postoperative complication, a wound infection.

PLPs were observed in five patients (four with Meckel's diverticulum and one duplication). Of these five patients, four experienced reduction failure, three had suspicious lesions on preoperative ultrasound or CT, and a PLP was discovered intraoperatively in one patient. One patient underwent surgery because a PLP was suspected after a successful air enema reduction. The presence of a PLP was not associated with age or recurrence, but the reduction success rate was significantly lower for the patients with PLPs (p < 0.001; Table 4).

3. Discussion

The recurrence rate in intussusception cases has varied greatly between series, with reported rates of 2–20% [9,11,12,16]. In the present

of recurrences. Among the 491 patients in our study, five had PLPs. The incidence of PLPs in children with intussusception has been reported to be 1.5–12% [20]. Many of the studies that reported PLPs in intussusception included cases of ileo-ileal and colo-colonic intussusception; these conditions are less frequent, but PLPs are more likely [3,22]. Our study included only ICI, which is considered to have a relatively low rate of PLPs.

In the present study, the patients with PLPs were not associated with recurrence, but the reduction success rate for these patients was significantly lower (P < 0.001). A recent large-scale study by Shen et al. reported that pathologic factors were independent risk factors for the failure of reduction [23]. The rate of pathologic intussusception was significantly higher in their failed reduction group than in the successful reduction group (28% vs. 0.8%). Other studies have also shown that the presence of pathologic factors was not associated with the recurrence of intussusception but was a main cause of the failure of reduction [15,24]. However, although only a minority of patients with recurrence

Table 3
Numbers and operation cases of RICI.

Recurrent attack	No. of cases	Recurrent episodes (early/late)	Operation case (n)	
1 recurrence	44	44 (17/27)	1	op at 1st recurrence. PLP(+)[*]
2 recurrences	15	30 (14/16)	1	op at 1st recurrence. PLP(-), 1 recurrence after operation
3 recurrences	3	9 (6/3)	1	op at 2nd recurrence. PLP(-), 1 recurrence after operation
4 recurrences	2	8 (4/4)	1	op at 4th recurrence. PLP(+)[*]
5 recurrences	3	15 (11/4)		
7 recurrences	1	7 (0/7)	1	op at 1st episode. PLP(-), 7 recurrences after operation

RICI ; recurrent ileocolic intussusception, op;operation, PLP; pathologic lead point.
 [*] Meckel's diverticulum.

study, the ICI recurrence rate was 13.8%, similar to the rates previously reported. The most appropriate treatment for RICI has not yet been established. In the past, many authors were in favor of surgical interventions for patients who experienced more than two episodes of recurrence to rule out the possibility of a PLP [17,18]. Some investigators have recommended surgical management after more than one recurrence in patients older than 2 years [19,20]. However, there are little data in support of a specific number of recurrences at which surgical intervention is recommended, or about whether children who experience multiple recurrences are at a greater risk of harboring a PLP or ischemic bowel, experiencing perforation at air enema, or subsequent recurrence.

Fisher et al. reported similar rates of PLPs among patients with one, two, or three or more episodes of intussusception [15], and Esmaeili-Dooki et al. suggested there was no association between recurrent intussusceptions and PLPs [16]. Conversely, some studies have reported that multiple recurrences were associated with a higher incidence of PLPs [9,21]. In the present study, the rate of PLPs did not differ between the recurrence and non-recurrence group, and there was no statistically significant relationship between the presence of a PLP and the number

Table 4
Comparison between PLPs groups and groups without PLPs.

	No PLP (n = 486)	PLP (n = 5)	P value
1st episode age			0.11
<1 y (n)	125	3	
≥1 y (n)	361	2	
Sex (M:F)	310:176	3:2	1.0
Reduction success rate (%)[*]	96.7	20	<0.001
Success (n)	470	1	
Fail (n)	16	4	
Operation (n)	15	5	<0.001
Bowel resection (n)	2	5	0.002
Recurrence case (n)	66	2	0.14

PLP; pathologic lead point.
 [*] air enema reduction.

will have PLPs, any recurrence should prompt a more vigorous investigation to exclude a PLP. In the present study, one patient who had undergone three reductions underwent surgery when a PLP was suspected on ultrasound and CT. We acquired CT scans for patients who experience multiple recurrences; we also performed a Meckel's scan for some patients.

Some previous reports have suggested there may be an association between the recurrence of intussusception and the patient's age, but this remains controversial. Some studies showed that patients with recurrent intussusception were younger than 1 year at the first episode, suggesting that this was because younger children may be more sensitive to viral triggers such as viral gastroenteritis [16]. Another study reported that intussusception patients older than 1 year tended to have higher early recurrence rates because older children were less likely to require operative reduction [14]. In the present study, there was no difference in recurrence rate and whether the recurrence was early or late between the patients younger than 1 year and those older than 1 year. As shown in Table 1, there was no significant difference in recurrence rate between the age subdivisions.

Arguments given for surgical intervention rather than air enema include the lower recurrence rate and a concern about reduced bowel viability after air enema. However, several studies have reported that the risks of surgical intervention and resection were lower for cases of recurrent intussusception than for a primary episode [11,25]. Daneman et al. and Justice et al. suggested that this was due to the shorter interval between the onset of recurrent symptoms and hospital visit in children who had experienced a previous intussusception, which probably reflects a greater awareness by parents of the clinical significance of the symptoms [11,25]. In the present study, no relationship was observed between recurrence and an increased rate of resection. Seven patients (five with PLPs) underwent bowel resection, and only two patients showed loss of intestine viability. Compared with air enema, the rate of recurrence was lower after operative reduction and was much lower after resection [15]. Despite this lower recurrence rate, surgery

did not ensure there was no recurrence. Among our patients, three patients developed recurrence after operative reduction; all were successfully treated with air enema.

In this series, there was no pattern of recurrence that was predictive of PLPs or the requirement for surgery.

Early and late recurrence episodes were not statistically associated with age, the presence of a PLP, or operation. Of the patients who experienced two or more recurrences, seven experienced only early recurrences, seven experienced only late recurrences, and 11 experienced both early and late recurrences. We therefore believe that each recurrence episode should be judged individually, regardless of whether it was early or late, and that the number of recurrences and whether they are early or late should not alter the treatment modality.

This study had its limitations. First, it had a single-center retrospective design, which may have resulted in errors in data collection and processing. Second, there was a potential for selection bias, because some of the cases with PLPs were not detected during the observation period, some of the patients with relatively short follow-up periods were included and screening to detect PLPs such as CT or Meckel's scan could not be performed in all patients. Finally, the sample size and the number of PLPs were small. Statistically, there was no significant difference between PLPs and recurrence, however, due to the small number, clinical conclusions are difficult to draw completely. Large-size multi-center studies are warranted. Despite these limitations, we expect that these data may help inform surgeons' decisions on whether to operate for RICI.

4. Conclusions

We suggest that the initial management of RICI should be by nonoperative treatment, regardless of which treatment they have previously received. Each recurrence should be managed as if it were the first episode. Operative reduction should be considered when nonoperative reduction fails, the presence of a PLP is suspected, or there are signs of peritonitis. Multiple recurrences are not a contraindication to air enema.

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