

Risk Factors for Recurrent Intussusception in Children: A Record Based Descriptive Study

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ABSTRACT

Introduction: Early diagnosis and management of cases of recurrent intussusception is challenging. It is well accepted that Pathological Lead Points (PLP) can precipitate recurrence. A few recent studies have identified association of a few clinical variables with recurrence.

Aim: To assess the frequency of clinical features and PLP in recurrent intussusception and to find the risk factors that lead to recurrence.

Materials and Methods: This was a retrospective cohort study that included medical records and ultrasound scan registry of children with intussusceptions treated with ultrasound guided hydrostatic saline reduction or surgery, between January 2010 to December 2017 in a Government Tertiary care and teaching institution. Totally, 362 cases of intussusceptions were reviewed.

Follow-up data were collected. There were 86 recurrent cases: 64 had single and 22 had multiple recurrences. The clinical parameters-age of child, gender, duration of symptoms, presence of vomiting, bloody stools, abdominal mass, ultrasound features, PLP identified, were evaluated. Fisher's-exact test and logistic regression analysis were used to measure significant factors affecting recurrent intussusception.

Results: After comparing recurrent and non-recurrent cases, it was determined that factors-age >1 year, shorter duration of symptoms (≤ 24 hrs), absence of vomiting and absence of bloody stools were significantly and independently associated with recurrent intussusception.

Conclusion: Intussusception in older children with shorter duration of symptoms and subtle clinical features need to be followed-up closely for recurrence.

Keywords: Bloody stools, Hydrostatic saline reduction, Pathological lead points

INTRODUCTION

Intussusception is a common abdominal emergency in infants and children [1]. Intussusception requires prompt diagnosis and management as this can lead to severe consequences including bowel infarction, perforation, peritonitis, and even death, if left untreated. Once intussusception has been diagnosed, air enema, barium enema or sonography guided hydrostatic reduction is used to reduce it with an excellent outcome [2,3].

Intussusceptions are commonly idiopathic but may be associated with Meckel's diverticulum, duplication, polyps etc. Also causes of multifocal bowel wall thickening like Henoch- Schoenlein purpura, cystic fibrosis, disordered coagulation, haemophilia lead to intussusception. Presence of pathologic lead points is seen in 0.3% to 20% of cases of intussusception [4].

Recurrent intussusception is defined as an occurrence of intussusception of a bowel loop in a patient with prior resolution of intussusception [5]. This can recur after a few hours, days, weeks or months after successful reduction. Recurrence of intussusception can occur regardless of the method of reduction [6]. The recurrence rate ranges from 8%-15% of intussusception after successful barium enema reduction [5], 5.2%-20% following sonography guided hydrostatic enema reduction [7] and 5.4%-15.4% following fluoroscopy guided air enema reduction [8].

Recurrent intussusceptions can usually be managed by repeat air enema, barium or sonographic hydrostatic saline reduction. Surgery is only used for irreducible recurrences, perforation, clinical or radiological evidence of PLP [8].

Early diagnosis and management of recurrence is challenging for paediatricians, paediatric surgeons and radiologists. PLP can precipitate recurrent intussusception, but reliable risk factors for most cases of recurrence have not been adequately identified. Few recent studies conducted in Chinese and Korean population have identified relationship between clinical parameters like age, symptom duration, vomiting and bloody stools with recurrence

[9,10]. Such patients can be closely followed-up for recurrence and appropriately managed.

The aim of the study was to assess the frequency of clinical features and PLP in recurrent intussusception and to find the risk factors that lead to recurrence. Therefore, a retrospective cohort study was performed to determine risk factors for recurrence of intussusception.

MATERIALS AND METHODS

The medical records and ultrasound scan registry data of 362 paediatric cases of intussusception at Government TD Medical College Alappuzha, Kerala, India, between January 2010 and December 2017 were analysed. This study was approved by Institutional Research and Ethics committee review board (No. B3/1573(A)/2010/TDMCA; EC48/2019).

Data was collected from case records in which final diagnoses of intussusception were confirmed by abdominal ultrasound scan, and saline hydrostatic reduction was performed. For all these patients, saline enema was performed at a height of 100-120 cm from procedure table under ultrasound guidance after informed consent, as per the institutional treatment protocol [11]. Surgery was found to be done in cases where reduction by pressure enema failed, signs of perforation or hemodynamic compromise was present or if there was a suspicious PLP. Cases of transient intussusception, small bowel intussusception and cases of spontaneous reduction were excluded from study. The data regarding clinical, ultrasound findings, PLP and per operative findings were noted. The follow-up data of these patients were also collected.

Recurrent intussusception was defined as recurrence of intussusception after sonographic guided hydrostatic saline reduction or surgery. Single recurrent intussusception was defined as cases that recurred only once after successful reduction. Multiple recurrent intussusception was defined as cases that recurred at least twice after saline reduction. Non-recurrent intussusception was defined as cases that were successfully reduced and has not recurred [9].

Thus cases were separated into two groups- Non-recurrent and Recurrent. The demographic factors were compared between the groups. They were grouped according to age (≤ 1 year and >1 year), gender, symptom duration (≤ 24 hrs and >24 hrs), symptoms (bloody stools, vomiting, location of abdominal mass), PLP identified, and were analysed. Cases of non-recurrent intussusception were used as control group when analysing the risk factors for recurrent intussusception. Follow-up data of all cases for a minimum period of at least 18 months after first episode of intussusception was collected.

STATISTICAL ANALYSIS

Data was presented as numbers and percentages. Univariate analysis of recurrent and non-recurrent group was done using Fisher's-exact test. Factors that contribute to recurrent intussusception were analysed using logistic regression analysis with SPSS statistics 21.0 (SPSS Inc., Chicago, IL, USA). The p-value <0.05 was considered to be statistically significant.

RESULTS

Clinical Characteristics of Recurrent Intussusception Cases

Among 362 patients, 196 patients (54.14%) were males and 166 (45.86%) were females. Recurrent intussusception was identified in 86 patients (23.7%).

In recurrent group, 58 (67.4%) were of age >1 year [Table/Fig-1]. The median age of recurrent group was 21 months. Among the recurrent cases, 64 were single recurrences, 12 had two recurrences, 8 had three recurrences, 2 had four or more recurrences; 8 (9.3%) cases of PLP were identified in recurrent group, and 10 (3.6%) in non-recurrent group.

Age	Recurrent male (%)	Recurrent female (%)	Non-recurrent male (%)	Non-recurrent female (%)
≤ 1 year	15 (4.1)	13 (3.6)	68 (18.8)	60 (16.6)
>1 year	32 (8.9)	26 (7.2)	81 (22.4)	67 (18.4)
Total	47 (13)	39 (10.8)	149 (41.2)	127 (35)

[Table/Fig-1]: Age and sex distribution in recurrent and non-recurrent group.

Predictors of Recurrence

[Table/Fig-2] shows the comparison of clinical characteristics of recurrent and non-recurrent cases of intussusception. Using univariate analysis, factors associated with recurrent intussusception were age >1 year, symptom duration ≤ 24 hours, absence of bloody stools, absence of vomiting, and presence of PLP ($p < 0.05$). There was no statistical significance in gender and right sided abdominal mass between two groups ($p > 0.05$).

Variables	Recurrent (n=86) (%)	Non-recurrent (n=276) (%)	χ^2 value	p-value
Male gender	47 (54.6)	149 (53.9)	0.0117	0.9139
Age >1 year	58 (67.4)	148 (53.6)	5.1057	0.0238
Duration of symptoms ≤ 24 hrs	72 (83.7)	55 (19.9)	117.16	<0.0001
Vomiting	20 (23.3)	190 (68.8)	44.75	<0.0001
Bloody stools	6 (6.9)	100 (36.2)	27.1	<0.0001
Right sided abdominal mass	85 (98.8)	270 (97.8)	0.3535	0.552
Pathological lead points identified	8 (9.3)	10 (3.6)	4.4756	0.0458

[Table/Fig-2]: Comparison of recurrent with non-recurrent intussusceptions.

Logistic regression analysis was performed to assess the independent predictors of recurrence among 362 patients. Age >1 year, symptom duration ≤ 24 hours, absence of bloody stools and absence of vomiting were significantly predictive of recurrent intussusception [Table/Fig-3].

Variables	OR	95% CI	p-value
Age >1 year	1.7915	1.0765 to 2.9813	0.0248
Duration of symptoms ≤ 24 hrs	20.6649	10.8509 to 39.3554	<0.0001
Vomiting	0.1372	0.07824 to 0.2405	<0.0001
Bloody stools	0.132	0.05557 to 0.3136	<0.0001

[Table/Fig-3]: Stepwise regression model of risk factors for recurrent intussusception.

Patterns of Recurrence

The following data was found for pattern of recurrences- within 24 hours of first reduction, there were 18 patients, between 24 hours and 1 week there were 34 patients, between 1 week and 1 month there were 14 patients, between 1 month and 6 months there were 12 patients, between 6 months and 1 year there were 6 patients and beyond 1 year there were 2 patients. The early recurrence rate (recurrence rate within 24 hrs) was 20.9%.

[Table/Fig-4] shows clinical characteristics and number of recurrences in recurrent intussusception cases. Single and multiple recurrent groups were again compared using univariate analysis. No significant difference was found in symptom duration, male gender, absence of bloody stools, absence of vomiting and right-sided abdominal mass. However, age >1 year was found to be statistically significant.

Variables	Single recurrence (n=64)	Multiple recurrence (n=22)	χ^2 value	p-value
Male gender	34	13	0.0539	0.8164
Age >1 year	38	20	7.413	0.006
Duration of symptoms ≤ 24 hrs	56	16	2.621	0.105
Vomiting	12	8	2.84	0.1413
Bloody stools	4	2	0.2035	0.6432
Right sided abdominal mass	63	22	0.3478	0.5554

[Table/Fig-4]: Comparison of single and multiple recurrent intussusceptions.

DISCUSSION

In present study, recurrence rate was 23.7%. Previous studies have noted that rate of recurrence of intussusception after non-surgical reduction ranges from 5-20% [7,10,12]. Lee DH et al., reported recurrence rate of 16.7% in their study [10]. Guo WL et al., conducted five-year retrospective study and found recurrence rate of 23.4% [9]. The lower rate of recurrence in many previous studies could be due to a higher rate of surgical reduction in them. Recurrence rate after manual operative reduction is usually only 1-3% [8,13]. In the present study, among 86 recurrent cases, only 19 underwent surgery. PLP were identified in eight cases. Most recurrences in the present study were treated by repeat saline reduction unless suspicious lead points, complications or three or more recurrences occurred.

Factors for Predicting Recurrent Intussusception

Patients older than one year age at the first episode of intussusception are more likely to experience recurrence. In this study, 67.4% of recurrent cases were aged more than one year. Xie X et al., and Shen G et al., reported age more than 2 years was risk factor for recurrence of intussusception [14,15]. Guo WL et al., found that 67.5% of recurrent cases were more than one year age [9]. This corresponds to the present study.

Male gender was not a significant risk factor for recurrence in this study. In a meta-analysis by Ye X et al., no association was found with gender and recurrence of intussusception [16],

In the present study, shorter duration of symptoms (≤ 24 hours) at first episode was found to be significant risk factor for recurrence. Champoux AN et al., had concluded that patients with recurrence have fewer symptoms and shorter duration [17]. Guo WL et al., also concluded that duration of symptoms <12 hours was risk factor for recurrence [9].

Absence of vomiting was a significant risk factor for recurrence in this study. This corresponds to Guo WL et al., where only 19% recurrent cases had vomiting [9]. This also corresponds to meta-analysis by Ye X et al., [16].

In this study, bloody stools were seen in 6% of recurrences. Absence of bloody stools was significantly associated with recurrence. In study by Guo WL et al., bloody stools were present in 4.7% of recurrent cases and absence of bloody stools was found to be a risk factor for recurrence [9]. This also corresponds to study by Ksia A et al., [12]. But in a study by Xie X et al., there was a contrary conclusion [14]. In meta-analysis by Ye X et al., bloody stools failed to show significant association with recurrence [16].

Right-sided abdominal mass was not found significantly associated with recurrence in the present study. In study by Guo WL et al., right-sided abdominal mass was found predictive of recurrence [9]. On the contrary, meta-analysis by Ye X et al., found no association between recurrent intussusception and location of mass [16].

Presence of PLP was statistically significant in recurrent cases in our study. In study by Guo WL et al., PLP was detected in 9.9% of recurrences and was found independent risk factor for recurrence [9]. But in the study by Fisher JG et al., recurrence did not predict PLP [18].

Patterns of Recurrence

In this study, the majority of recurrences were single recurrences (74.4%). Also, early recurrence rate (recurrence within 24 hrs) was 20.9%. Univariate analysis showed there were no significant differences in male gender, symptom duration, bloody stool, vomiting between single recurrence and multiple recurrence intussusception groups. In study by Guo WL et al., the percentage of single recurrence was 45.5% of total recurrences and early recurrence rate was 6.2% [9]. Thus it is very important to observe such patients during the early period after hydrostatic reduction.

LIMITATION

The greatest limitation of our study was the retrospective design and the conclusions were based on observation from a single centre. Also, the sample size was relatively low. Thus, a prospective multicentre study is required to further evaluate the predictors of recurrence.

CONCLUSION

When older children with subtle clinical features present with first episode of intussusception, they should be watched more

closely for recurrence. Parents of such children should be warned sufficiently about possible recurrence and need for immediate further medical attention.

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