



Clinical characteristics and surgical outcome in children with intussusceptions secondary to pathologic lead points: retrospective study in a single institution

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Abstract

Background Intussusception secondary to pathologic lead points (PLPs) is a potential surgical emergency and almost all cases need surgery. The aim of this study was to evaluate the clinical manifestations, physical examinations and surgical outcomes of secondary intussusception (SI) caused by PLPs, as well as to improve the diagnosis and treatment of PLPs in children and infants.

Materials and methods We retrospectively reviewed the records of 83 children and infants who were diagnosed with intussusception secondary to PLPs in our institution. The ultimate diagnosis was dependent on histopathological findings under a microscope by a pathologist. Patients were divided into a younger group (< 2 years old) and the older group (> 2 years old) according to age. Patient demographics, clinical manifestations, duration of symptoms, auxiliary examinations, and the presence of pathological lead point were recorded.

Results A total of 83 patients were found with intussusception secondary to PLPs in this study. Patients were aged from 4 days to 14 years, with a mean age of 3.8 years (median 1.5; range 0–14 years). There were 47 cases in the younger group and 36 cases in the older group. The main clinical symptoms were intermittent crying or abdominal pain. PLPs were observed in only ten patients on US (12%). Ten patients underwent enteroscopy examination for further diagnosis, and all the patients had positive findings including seven cases of Peutz–Jeghers syndrome and three cases of benign polyps. Technetium-99 m pertechnetate scans were performed in ten patients and five patients had positive results (50%). Based on the surgical findings, complex/compound is the most common type of intussusception, followed by small intestinal and ileo-colic type. The main types of PLPs were Meckel's diverticulum ($n = 31$), duplication cyst ($n = 19$) and benign polyps ($n = 13$). Meckel's diverticulum and intestinal duplication were the most common causes of secondary intussusception among children younger than 2 years, accounting for 81% (38/47) of the cases. The most common causes of secondary intussusception in children older than 2 years were intestinal polyps, Meckel's diverticulum and Peutz–Jeghers syndrome, accounting for 72% (26/36) of the cases.

Conclusions The presence of a pathological lead point is more likely in older children. The most common types of intussusception secondary to PLPs are complex/compound and small intestinal. Meckel's diverticulum and intestinal duplication were the most common causes of secondary intussusception among younger children and Peutz–Jeghers syndrome and intestinal polyps were commonly seen in older children.

Keywords Intussusception · Pathologic lead points · Pediatrics · Diagnosis

Lingling Zhao, Shaoguang Feng, Peng Wu and Xin-He Lai contributed equally to the work.

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Introduction

Intussusception is the most common cause of intestinal obstruction and abdominal emergency in young children [1–3]. Timely diagnosis and treatment are important, as any delay may lead to serious complications, such as intestinal perforation, bowel resection and mortality [4]. Hydrostatic or pneumatic reduction has become the first-line treatment

for the majority of cases with high success rate [5]. Operative reduction is needed only if reduction failed, suspected of pathological causes or intestinal necrosis or perforation. Most intussusceptions were idiopathic, and secondary intussusception caused by PLPs was found in 2.2–15% of childhood [6–8]. For those SIs caused by PLPs, surgical treatment is ultimately required. The presence of a PLP is more likely in older children [3]. Operative intervention should not be delayed in these patients if difficult or doubtful pneumatic reduction is encountered, especially in those older children. If not timely managed, it can cause some serious complications, such as bowel perforation and secondary peritonitis, which can result in death in extreme cases. Therefore, early diagnosis of SI is very important.

The aims of this study were to summarize the clinical manifestations, physical examinations and surgical outcomes of secondary intussusception caused by PLPs, as well as to improve the diagnosis and treatment of PLPs.

Materials and methods

After obtaining institutional review board approval, we performed a retrospective review of 83 children who were diagnosed with intussusception secondary to PLPs in our institution between January 2002 and December 2018. The preliminary diagnosis of a secondary intussusception was made by the surgeon on the basis of gross appearance, while the ultimate diagnosis was dependent on histopathological findings under a microscope by a pathologist. There were five different types of intussusceptions based on the anatomic location: the definition of small intestinal intussusception is proximal small intestinal segment prolapsed into the adjacent distal small intestine; ileo-cecal and ileo-colic defined as one distal ileum segment prolapsed into adjacent cecum and adjacent colon, respectively; definition of complex/compound is more than one proximal intestinal

segment prolapsed into the adjacent one, and multiple defined as more than one intussusception in various intestinal segments. Only pediatric patients under 14 years old were included in this study. Patients were divided into the younger group (<2 years old) and the older group (>2 years old) according to age. Patient demographics, clinical manifestations, duration of symptoms, auxiliary examinations, and the presence of pathological lead points were recorded.

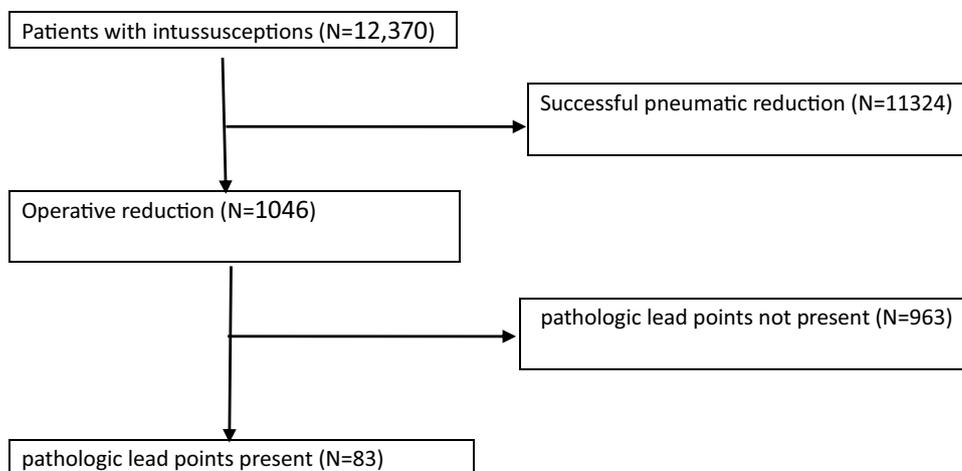
Statistical analysis

SPSS Version 20.0 was applied for statistical analysis. All continuous variables were tested for normality using the Kolmogorov–Smirnov test. For normal distributed data, Student's *t* test was used to compare the groups. For non-normal data distribution, the Kruskal–Wallis test was used instead. Comparison between groups was performed using Pearson's chi-square test for categorical variables. Results were considered statistically significant if analysis yielded a *p* value <0.05.

Results

A total of 12,370 pediatric patients with intussusception were admitted to our hospital. Most of the patients without evidence of shock or peritonitis had successful pneumatic reductions and only 1046 cases underwent surgery. Finally, a total of 83 patients were found with intussusception secondary to PLPs in this study (Fig. 1). Patients were aged from 4 days to 14 years, with a mean age of 3.8 years (median 1.5; range 0–14 years). There were 47 cases in the younger group and 36 cases in the older group. The main clinical symptoms were intermittent crying or abdominal pain. Vomiting occurred in 40 patients (48.2%). Thirty-nine patients (47.0%) had bloody stool and there were significantly more bloody stools in the younger group ($p=0.009$) than in the

Fig. 1 Study flow of the treatment of our group of patients with intussusception



older group. The duration of symptoms ranged from several hours to 3 days. There was no significant difference in the duration of symptoms between the two groups. Forty-eight (57.8%) had typical signs of a palpable mass in the abdomen (Tables 1, 2).

Ultrasonography (US) was performed in all patients with intussusception secondary to PLPs, and intussusception was diagnosed by US. However, PLPs were observed in only ten patients on US (12%). Ten patients underwent enteroscopy examination for further diagnosis, and all the patients had positive findings including seven cases of Peutz–Jeghers syndrome and three cases of benign polyps. Technetium-99 m pertechnetate scans were performed in ten patients and five patients had positive results (50%) (Table 1). Based on the surgical findings, all cases of PLPs intussusception were divided into five different types on the basis of the anatomic location of intussusception: small intestinal, ileo-colic, ileo-cecal, complex/compound and multiple. Complex/compound was the most common type of intussusception, followed by small intestinal and ileo-colic type. The types of intussusceptions are shown in Table 3.

Table 1 Descriptive data for intussusception secondary to pathologic lead points (PLPs)

Variable	Data
Gender, <i>n</i> (%)	
Male	53 (63.9%)
Female	30 (36.1%)
Age (years)	
< 2	47 (56.6%)
> 2	36 (43.4%)
Clinical manifestations	
Abdominal pain	45 (54.2%)
Crying	56 (67.5%)
Vomiting	40 (48.2%)
Palpable mass	48 (57.8%)
Bloody stool	39 (47.0%)
Mean time to surgery [range], h	27.8 [3–96]
Recurrent intussusception	42 (50.6%)
Auxiliary examinations, <i>n</i> (%)	
US	10 (30%)
Enteroscopy	10 (100%)
ECT	5 (50%)

Table 2 Time to intervention and clinical features of the patients with PLPs between the groups

Variable	Young (< 2 years)	Old (> 2 years)	<i>p</i> value
Mean time to intervention, h	28.6 ± 24.5	26.7 ± 24.9	0.54
Recurrent intussusception	22	20	0.43
Bloody stool	29	10	0.002
Suspected pathological causes	12	18	0.021

Table 3 Distribution of the types of intussusception in patients between the groups

PLPs	Young (< 2 years)	Old (> 2 years)	<i>p</i> value
Ileo-colic	11	8	0.899
Ileo-cecal	5	4	0.945
Complex/compound	18	16	0.573
Small intestinal	12	8	0.727
Multiple	1	0	0.379

The main types of PLPs were Meckel diverticulum (*n* = 31), duplication cyst (*n* = 19) and benign polyps (*n* = 13). Other less frequent PLPs included eight Peutz–Jeghers syndrome, five mesenteric cysts, three malignant lymphomas, three allergic purpura and one hamartoma. Meckel’s diverticulum and intestinal duplication were the most common causes of secondary intussusception among children younger than 2 years, accounting for 81% (38/47) of the cases. The most common causes of secondary intussusception in children older than 2 years were intestinal polyps, Meckel’s diverticulum and Peutz–Jeghers syndrome, accounting for 72% (26/36) of the cases (Table 4).

Discussion

Intussusception is a gastrointestinal disorder describing the involution of a proximal portion of the bowel into a more distal portion, often causing intestinal inflammation and bowel obstruction. The vast majority of intussusception cases are idiopathic with good prognosis; most of previous studies have focused on these cases. A few cases of intussusception are caused by pathological lead points, such as Meckel’s diverticulum, duplication, polyp and tumors, which are relatively difficult to diagnose and confirm before surgery. It is rather important to explore the primary disease of PLPs because PLPs could lead to repeated unnecessary enemas and sometimes poor prognosis [9]. With improved technical instruments, more and more PLPs could be identified and treated at an early stage, for example, the Technetium-99 m pertechnetate scan is the most useful method for the detection of Meckel’s diverticulum with high sensitivity and specificity [10]. Enteroscopy is the best diagnostic method

Table 4 Distribution of PLPs in patients with intussusception between the groups

PLPs	Young (<2 years)	Old (>2 years)	<i>p</i> value
Meckel diverticulum	22	9	0.042
Duplication cyst	16	3	0.006
Benign polyps	3	10	0.008
Malignant lymphoma	0	3	0.044
Mesenteric cysts	4	1	0.277
Peutz–Jeghers syndrome	1	7	0.008
Allergic purpura	1	2	0.407
Hamartoma	0	1	0.250

for the children with intestinal polyps such as Peutz–Jeghers syndrome. Despite all this, the majority of PLPs cases are diagnosed during operation.

Our study revealed that there were more male patients than female patients, with a male:female ratio of 5.3:3, which was similar with the previous report [11]. Lin et al [12] reported that the ratio of male:female is 4:1. Although our study shown that PLPs occurred in children older than 2 years were similar with children younger than 2 years, most intussusceptions occur in children younger than 1 year and almost all intussusceptions occurred in children older than 5 years old are secondary to PLPs [13]. Therefore, the incidence of intussusception secondary to PLPs is much higher in children older than 2 years compared to children younger than 2 years, although the total number is similar. Clinical presentations of intussusception secondary to PLPs were lack of specificity, which were similar to primary intussusception. In general, the classic symptoms were emesis, intermittent abdominal pain and bloody stools with or without a palpable mass in the abdomen [14]. The types of intussusception of primary intussusception and secondary intussusception are also different. Most types of primary intussusceptions are ileo-colic, while intussusception secondary to PLPs could be complex/compound (such as ileoileal and ileoileocolic), small intestinal, or colocolic [15]. As demonstrated in the current study, the most common types of intussusception secondary to PLPs are complex/compound and small intestinal.

Pre-operative auxiliary examinations such as ultrasound and CT not only can help the diagnosis of intussusception, but also may search for a lead point [16, 17]. Ultrasonic diagnosis depends highly on the skills of physicians. In the study by Zhang et al. [15], among the 37 secondary intussusception patients, 21 cases (56.8%) were accurately categorized with lesions, including intestinal polyps, cystic intestinal duplication, intestinal wall lymphoma, and a small part of Meckel's diverticulum. In our series, all intussusception cases were detected by US. However, PLPs were observed

in only ten patients on US. In our study, ten children with recurrent intussusception underwent a Technetium-99 m pertechnetate scan and five (50%) patients were diagnosed PLPs prospectively. The most common causes of secondary intussusception among all children are Meckel's diverticulum and intestinal duplication. Other rare causes include intestinal polyps, allergic purpura, Peutz–Jeghers syndrome and intestinal tumors [3, 12, 18, 19]. Our current study was consistent with previous reports. On further analysis, the most common causes of secondary intussusception among children younger than 2 years were Meckel's diverticulum and intestinal duplication, which accounted for 81% (38/47) of the cases, while in children older than 2 years intestinal polyps, Meckel's diverticulum and Peutz–Jeghers syndrome were the most common causes of secondary intussusception, accounting for 72% (26/36) of the cases.

In conclusion, based on the results of our analyses on the 83 pediatric patients with intussusception secondary to PLPs that underwent surgery, we conclude that the incidence of a pathological lead point is more likely in older children. The most common types of intussusception secondary to PLPs are complex/compound and small intestinal. Meckel's diverticulum and intestinal duplication were the most common causes of secondary intussusception among younger children, and Peutz–Jeghers syndrome and intestinal polyps were commonly seen in older children.

Compliance with ethical standards

Conflict of interest The authors report no conflicts of interest in this work.

Human rights and animal participants This article does not contain any studies with human participants or animals performed by any of the authors.

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