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### Operative indications in recurrent ileocolic intussusception

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ABSTRACT

Background: Air-contrast enema (ACE) is standard treatment for primary ileocolic intussusception. Management of recurrences is less clear. This study aimed to delineate appropriate therapy by quantifying the relationship between recurrence and need for bowel resection, pathologic lead points (PLP), and complication rates. *Methods:* After IRB approval, a single institution review of patients with ileocolic intussusception from 1997 to 2013 was performed, noting recurrences, outcomes, and complications. Fisher's exact and t-tests were used. *Results:* Of 716 intussusceptions, 666 were ileocecal. Forty-four underwent bowel resection, with 29 PLPs and 9 ischemia/perforation. Recurrence after ACE occurred in 96 (14%). Recurrence did not predict PLP (P = 0.25). Recurrence (≥ 3) was associated with higher resection rate (P = 0.03), but not ischemia/perforation (P = 0.75). ACE-related complications occurred in 4 (0.5%) patients. Successful initial ACE had 98% negative predictive value for resection, and PLP (e.g., after successful ACE, 2% had resections, 2% PLP). After failed initial ACE, 36% received resection, and 23% had PLP (P < 0.001). *Conclusions:* Recurrence is associated with a greater risk of resection but not PLP or ACE-complication. Failed ACE is associated with increased risk for harboring PLP and receiving resection. ACE should be the standard treatment in recurrent intussusception, regardless of number of recurrences.

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lleocolic intussusception is the second most common cause of bowel obstruction in pediatric patients with at least 56 cases per 100,000 children per year in the United States [1]. Reduction of the intussusception by air-contrast enema (ACE) has a high success rate and is well tolerated with few complications [2]. For this reason, ACE is typically first line therapy for the initial episode of intussusception. Surgery is reserved for children with peritonitis, shock/sepsis, pneumoperitoneum, or a preoperatively evident pathologic lead point (PLP) [3].

However, intussusception recurs in 9%–18% of children after nonoperative reduction [4–9], and the indications for operation in recurrent intussusception are unclear [9–11]. Some authors have recommended surgical intervention after more than one [12] or two episodes [11,13,14]. Others contend that surgery should be reserved for children whose intussusceptions fail to reduce with ACE or contrast enema [5,15].

Since a randomized controlled trial demonstrated safety and efficacy of ACE while using lower doses of radiation compared with barium enema (BE) [16], ACE has become the standard of care for the initial episode of intussusception in North America [17,18]. Some data suggest that repeat enema for treatment of the first recurrence is safe [15,19,20] and ACE has been successfully used to treat multiple recurrences [5–7,9]. Much of the extant data regarding the management of recurrent intussusception, however, include patients who were treated with BE rather than ACE. Few studies exist that have evaluated the management and

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operative indications for children with recurrent intussusception solely in the era of ACE [7,9]. Since it appears that multiple recurrences can be treated safely and effectively with ACE, the surgical criteria are even less clear. Thus a contemporaneous study evaluating the need for surgical management in recurrent intussusception would be relevant and useful to the current management of this disease process.

This study therefore aimed to delineate operative indications for recurrent intussusception in the era of ACE. The specific goal was to determine if a certain number of recurrences should warrant operative intervention based on traditional indications for surgery: (1) concern for PLP, (2) concern for bowel damage requiring resection, (3), consideration of possible further recurrence, and (4) concern for increasing risk of an ACE-related complication. Hence, the impact of recurrence on each of these factors was assessed.

#### 1. Methods

Following approval from the institutional review board (IRB #P00008210), medical records from a single institution were retrospectively reviewed. Children 0–18 years of age with a diagnosis code for intussusception (ICD-9 560.0) between March 1997 and March 2013 were screened. Patients with ultrasound-proven intussusception were evaluated and only those with ileocolic intussusception were included for analysis. Data recorded included total number of intussusception episodes within the study period, ACE and operative procedure details and outcomes, as well as imaging and histopathologic findings. Each patient's record was specifically screened for history of operation,

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presence of PLP, intestinal ischemia, intestinal perforation, and intestinal resection.

ACEs were performed according to the institutional protocol using the commercially available Shiels intussusception air reduction system (Custom Medical Products, Maineville, OH) [21]. This device consists of a disposable tubing set with an enema tip and a reusable aneroid gauge and insufflator. The tip is inserted with a rubber disk gasket that fits at the anus. The apparatus is secured to the buttocks using cloth tape to ensure a tight seal. Under fluoroscopy, the colon is insufflated. The peak pressure applied is 120 mmHg in a calm child and may rise higher with crying or valsalva. Visible reduction of the intussusception and free flow of air into the small bowel are the criteria for successful treatment. The protocol is the same for both initial and recurrent intussusceptions.

Statistical analysis was completed using JMP (SAS Institute, Cary, NC). Continuous variables are presented as mean  $\pm$  standard deviation. Comparisons between groups were made using Student's t-test and Chi-squared test where appropriate. Rates of resection, PLP, and bowel ischemia/perforation were compared using Fisher's exact test. Sensitivities, specificities, and positive and negative predictive values were calculated based on failure of ACE for predicting PLP and receiving a resection. Predictive values were calculated using Bayesian conditional probability.

#### 2. Results

Of 716 confirmed cases of primary intussusception identified over the 16 year period, 50 involved only small bowel and none involved only colon. The remaining 666 were ileocolic and were included in this analysis. At the time of initial intussusception, 636 underwent ACE, 8 spontaneously reduced, and 22 underwent operations (for high degree of suspicion for PLP or bowel ischemia). Of these 666, 35 (5%) had operations after a successful ACE reduction due to a suggestion of PLP on ACE. A total of 96 patients had at least a single recurrence of intussusception after ACE reduction. Recurrences occurred at 2.7  $\pm$ 5.6 months (range <24 h to 30 months). Time to recurrence was not different between patients with PLPs (2.6  $\pm$  9.3) and without PLPs (4.4  $\pm$ 5.3, P = 0.45). Among the 6 patients with PLP and recurrence, the times to recurrence were: 0, 0, 1, 3, 92, and 697 days (23 months). Management and outcomes of patients for each episode of intussusception are shown in Fig. 1.

Comparing those with (n = 96) and without recurrence (n = 570), age was similar  $(2.1 \pm 1.3 \text{ versus } 2.1 \pm 2.3 \text{ years}, P = 0.38)$  and there was no difference in sex (69 versus 64% male, P = 0.33).

The primary outcomes regarding PLPs, bowel resection rates, and presence of ischemia or perforation are shown in Table 1. Only a single episode of intussusception was observed in 570 patients, while 65 had two episodes, 14 had three, 10 had four, 5 had five, 0 had six, and 2

#### Table 1

Bowel resections, pathologic lead points, and perforation or ischemia by episode.

	Single Episode (Percent)	2 Episodes	3 or more episodes	P value
Patients	570	65	31	
Resections	35 (6.1)	3 (4.6)	6 (19.4)	0.03 <sup>a</sup>
Pathologic lead points	23 (4.0)	3 (4.6)	3 (9.7)	0.25
Ischemia or perforation	9 (1.6)	0 (0)	0 (0)	-

Comparisons made using Fisher's exact test.

<sup>a</sup> The rate of resection was higher in patients with three or more episodes. There was no difference in resection rates between those with single and two episodes.

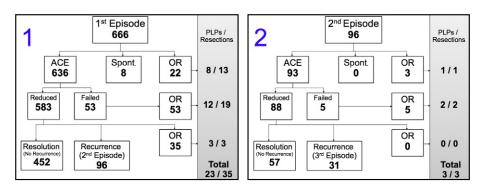
had seven episodes. Those with three or more episodes were combined for the purpose of analysis given the small numbers with further recurrences. The chance of encountering a PLP was not statistically different between one, two, and three or more episode groups (P = 0.25).

Children older than 5 years were more likely to have PLPs [13 of 45 (29%) versus 16 of 621 (3%), P < 0.001], accounting for a substantial portion of all PLPs found (13/29 = 45% despite only representing 45/666 = 7% of the study group). Of this group, 20 of 45 (44%) underwent an operation at the initial presentation. However, when stratified by age group ( $\leq$ 5 and >5 years), recurrence was still not a risk factor for PLP in both groups (P > 0.05). In addition, no other risk factors for PLP were identified in the lower age group. The PLPs encountered and the associated number of episodes are detailed in Table 2. The majority were Meckel's diverticuli (n = 18, 62%). Of note, 4 patients (14% of PLPs) were found to have B cell lymphomas.

Patients with three or more episodes were more likely to receive a bowel resection than those with one or two (P = 0.03). However, a pathological finding of tissue ischemia or perforation was only found in patients with a single episode of intussusception. Two of the 5 patients with 5 episodes underwent resections and did not have PLPs. In those cases, the indication for resection was surgeon concern for PLP (i.e. no gross evidence of ischemic damage). Of the 2 patients who had 7 episodes, 1 had a resection, which was performed to prevent further recurrence (no gross or histologic pathologic findings were seen).

The success or failure of ACE to reduce intussusception predicted the absence or presence of PLP and whether or not the patient received a bowel resection. Using ACE-failure as a test of PLP presence and of receiving a resection, the sensitivity, specificity, and positive and negative predictive values were calculated (Table 3). Both 1st ACE-failure and any ACE-failure were moderately sensitive and highly specific for PLP presence and performance of a resection. The negative predictive values of both 1st ACE-failure and any ACE-failure for both PLP and resection were all 98%. In other words, the chance of receiving a resection or finding a PLP after a successful ACE was 2%.

Following the first episode, there were 96 recurrences after 583 successful ACEs (16.5%). Recurrences after each additional episode are



**Fig. 1.** Flow chart for management of children with ileocolic intussusception through the second episode. Of those with 3 or more episodes (n = 31), there were 12 operations, 6 resections, and 3 PLPs. ACE = air contrast enema, Spont. = spontaneously reduced, PLP = pathologic lead point.

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