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Dutch Intussusception Grp; Kanglie, Maadrika M. N. P.; de Graaf, Nanko; Beije, Femke; Brouwers, Elise M. J.; Theuns-Valks, Sabine D. M.; Jansen, Frits H.; van Zuidewijn, Diederick B. W. de Roy; Verhoeven, Bas; Van Rijn, Rick R.

Published in:
Journal of Pediatric Surgery

DOI:
10.1016/j.jpedsurg.2018.05.006

Journal of Pediatric Surgery 54 (2019) 500–506

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

The incidence of negative intraoperative findings after unsuccessful hydrostatic reduction of ileocolic intussusception in children: A retrospective analysis☆

Maadrika M.N.P. Kanglie [a], Nanko de Graaf [b], Femke Beije [c], Elise M.J. Brouwers [d], Sabine D.M. Theuns-Valks [e], Frits H. Jansen [f], Diederick B.W. de Roy van Zuidewijn [g], Bas Verhoeven [h], Rick R. van Rijn [i], Roel Bakx [j,*], on behalf of the Dutch Intussusception Group

[a] Faculty of Medicine, University of Amsterdam, The Netherlands
[b] Department of Paediatric Radiology, Sophia Children's Hospital, Erasmus University Medical Centre, Rotterdam, The Netherlands
[c] Department of Emergency Medicine, Isala hospital, Zwolle, The Netherlands
[d] Department of Radiology, Albert Schweitzer Hospital, Dordrecht, The Netherlands
[e] Department of Paediatric Gastroenterology, Albert Schweitzer Hospital, Dordrecht, The Netherlands
[f] Department of Radiology, Catharina Hospital, Eindhoven, The Netherlands
[g] Department of Surgery, Medical Centre Leeuwarden, Leeuwarden, The Netherlands
[h] Department of Paediatric Surgery, Radboud University Medical Centre, Nijmegen, The Netherlands
[i] Department of Radiology, Emma Children's Hospital - Academic Medical Centre Amsterdam, The Netherlands
[j] Department of Paediatric Surgery, Emma Children's Hospital - Academic Medical Centre Amsterdam, The Netherlands

ARTICLE INFO

Article history:
Received 19 January 2018
Received in revised form 6 April 2018
Accepted 10 May 2018

Key words:
Intussusception
Hydrostatic reduction
Negative intraoperative findings
Pediatric surgery
Laparotomy
Laparoscopy

ABSTRACT

Background: There is a lack of studies addressing the occurrence of negative intraoperative findings (that is the absence of intussusception) after an unsuccessful hydrostatic reduction of an ileocolic intussusception. The aim of this study is to determine the incidence of negative intraoperative findings after unsuccessful hydrostatic reduction of ileocolic intussusception.
Methods: We conducted a multicentre retrospective study of all children aged 0–18 years treated for ileocolic intussusception from January 1, 2010 to December 31, 2015 in 9 Dutch hospitals. Primary outcome measure was the percentage of children without an intussusception during surgical exploration after unsuccessful hydrostatic reduction.
Results: In the study period 436 patients were diagnosed with an ileocolic intussusception. Of these, 408 patients underwent hydrostatic reduction of an ileocolic intussusception. 112 patients (27.5%) underwent surgery after an unsuccessful hydrostatic reduction. In 13 (11.6%) patients no intraoperative evidence of intussusception was found. Patients who underwent surgical intervention after unsuccessful hydrostatic reduction were significantly younger than patients who had a successful hydrostatic reduction; there was no gender difference.
Conclusion: A substantial number of children (11.6%) underwent a laparotomy after unsuccessful hydrostatic reduction in whom no intussusception was found intraoperatively. We suggest initiating laparoscopy instead of laparotomy when surgery is necessary.
Level of evidence: Level II.

In intussusception, a proximal segment of intestine invaginates into the adjoining distal intestinal lumen, leading to bowel obstruction [1]. It is a disease which mostly presents in pediatric age and is a common abdominal emergency in children. The classical presentation is that of a sick child with abdominal cramps, vomiting and blood in the diaper ("currant jelly stool") [2]. This classical presentation is, however, present in only up to 30% of cases [3,4]. Left untreated, intussusception could lead to the death of the child. The highest incidence of intussusception is in children under the age of 1 year with a decline in the following higher age categories [5,6].

Abbreviations: SN, Statistics Netherlands; DRG, Diagnosis Related Group registration; ICD, International Classification of Diseases; US, Ultrasonography; IRB, Internal Review Board; PA, Histopathological Analysis; IV, Intravenous.

☆ Declarations of interest: none.This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

* Corresponding author at: Department of Paediatric Surgery, Emma Children's Hospital - Academic Medical Centre Amsterdam, Meibergdreef 9, 1105, AZ, Amsterdam, The Netherlands. Tel.: +31 20 5669111.
E-mail address: r.bakx@amc.uva.nl (R. Bakx).

https://doi.org/10.1016/j.jpedsurg.2018.05.006
0022-3468/

According to a literature review conducted by the World Health Organization (WHO), the estimated mean annual incidence worldwide is 50–250 per 100.000 live births [7]. A Swiss study showed an incidence of 38, 31, and 26 cases per 100.000 children in respectively the first, second and third year of life [1].

According to Statistics Netherlands (SN) there were 3.429.193 Dutch citizens under the age of 18 years on 1–1-2015, of these 174.681 were under the age of 1 year [8]. Based on the Swiss data this would lead to an estimated number of 66, 54 and 47 children with intussusception in respectively the first, second and third year of life in 2015 in the Netherlands. The Dutch Healthcare Authority, based on diagnosis related group (DRG) registration, calculated that 290 children annually have an intussusception [9].

The choice of treatment for a clinically stable child with idiopathic ileocolic intussusception is either hydrostatic or pneumostatic reduction. If this is unsuccessful or if perforation is suspected, surgical reduction must be conducted. No literature specifically addressing the occurrence of negative intraoperative findings (defined as the absence of intussusception) after unsuccessful radiological reduction of ileocolic intussusceptions is available.

The aim of this study is to determine the incidence of negative intraoperative findings after unsuccessful radiological reduction of ileocolic intussusception in children in the Netherlands.

1. Material and methods

We conducted a retrospective multicentre study of children treated for an intussusception from January 2010 to December 2015 in all Dutch pediatric surgical centres: Emma Children's Hospital – Academic Medical Centre Amsterdam, Sophia Children's Hospital – Erasmus Medical Centre Rotterdam, Juliana Children's Hospital – Hagahospital the Hague, Maastricht University Medical Centre +, Radboud University Medical Centre Nijmegen, University Medical Centre Groningen, VU University Medical Centre Amsterdam, Wilhelmina Children's Hospital – University Medical Centre Utrecht, and 1 general hospital (Albert Schweitzer Hospital, Dordrecht).

Patients were identified using the International Classification of Diseases (ICD), book 10 code for intussusception, K56.1 or by searching on the DRG code for intussusception. All patients aged 0–18 years presenting with a suspected intussusception in one of the 9 participating hospitals were included. After applying the exclusion criteria, the final study population consisted of all children with an ileocolic intussusception who underwent radiological reduction. In those cases where a negative intraoperative finding was found the radiological studies were reviewed by a pediatric radiologist with 14 years' experience (RvR).

1.1. Study parameters

Clinical characteristics and medical histories were retrospectively retrieved from the medical records of each individual patient and were recorded in an anonymized database. The following characteristics were recorded: date of birth, gender, date of presentation, radiologic diagnosis, radiological reduction technique, cause of the intussusception, length of the intussusception, radiologist's experience level, surgical reduction technique, surgical findings, surgical intervention, time between failed radiological reduction and surgical reduction, recurrence rate, and length of hospital stay. The cause of the intussusception in the non-surgical group was based on imaging findings, as reported by the radiologist performing the ultrasound study, and in the surgical group on either the per-operative or pathology findings. A successful reduction was defined as a reduction where overflow of contrast into the terminal ileum was reported. An unsuccessful reduction was defined as a persistent intussusception after three reduction attempts in one single session. In the Netherlands radiologists are trained to try to reduce three times 5 min in one session. At the discretion of the treating team a second delayed reduction attempt was performed.

A recurrence was defined as an ileocolic intussusception within 30 days after the last successfully reduced intussusception. A patient with an ileocolic intussusception 30 days or more after the previous episode of a successfully reduced intussusception was recorded as a new patient.

1.2. Diagnosis

The diagnosis of intussusception was based on ultrasonography (US). Following a positive diagnosis children either underwent radiological reduction, surgical reduction or were referred to a specialized pediatric medical centre for radiological or surgical reduction.

1.3. Primary outcome measure

The primary outcome measure was the percentage of children with an ileocolic intussusception with negative intraoperative finding (that is the absence of intussusception), after an unsuccessful radiological reduction.

1.4. Secondary outcome measures

Three secondary outcome measures were defined: first, the success rate for radiological reduction in this Dutch Cohort; second, the distribution of surgical techniques and the percentage of cases in which a conversion from laparoscopy to laparotomy occurred; third and finally, the cause of the intussusception, defined as lesions at the lead point of the intussusception. For this we considered enlarged lymph nodes, lymphoid hyperplasia and Peyer's patches as idiopathic causes of intussusception, as these are variations of normal anatomy in response to an inflammation. The diagnosis of the cause of intussusception in non-operated patients and in operated patients with no specimens obtained during surgery was made based on sonography findings or by observations made during the operation. The diagnosis of the cause of the intussusception in operated patients with specimens obtained during surgery was made based on the histopathological analysis.

1.5. Statistical analysis

Differences between baseline characteristics were analyzed using a chi-square test or Mann–Whitney *U* test when appropriate. A 2-tailed $p < .05$ was chosen as the threshold for statistical significance. These and other descriptive analyses were performed using Statistical Package for Social Sciences v. 24 (IBM, Chicago, IL).

1.6. Internal review board approval

The internal review board (IRB) of the Academic Medical Centre Amsterdam issued a waiver for this retrospective study. Three hospitals (Albert Schweitzer Hospital, Erasmus University Medical Centre, and University Medical Centre Utrecht) required and obtained additional local IRB approval for the study.

2. Results

2.1. Patient characteristics

Between 2010 and 2015 593 patients were presented in one of the 9 participating medical centres with a suspected intussusception (baseline characteristics are reported in Table 1). Of these patients, 410 were boys (69.1%) and 183 were girls (30.9%). The median age of the whole study population was 1.7 years (IQR 2.7). The median age of the male patients was 1.6 years (IQR 2.6) and the median age of the female patients was 1.7 years (IQR 1.7). In 436 patients an ileocolic intussusception was diagnosed, of these, 2 patients were referred to a pediatric surgical centre (these were excluded to prevent duplication

Table 1
Baseline characteristics.

		Ileocolic Intussusception (N = 428)[a]			Final study group Hydrostatic Reduction (N = 408)[b]		
	Total population	Hydrostatic reduction	Surgical reduction	P[c]	Successful reduction	Unsuccessful reduction	P[c]
Number of Patients (%)	593	408 (93.6%)	20 (4.6%)		293 (71.8%)	112 (27.5%)	
Gender (%)							
Male	410 (69.1%)	282 (69.1%)	11 (55.0%)		199 (67.9%)	80 (71.4%)	
Female	183 (30.9%)	126 (30.9%)	9 (45.0%)		94 (32.1%)	32 (28.6%)	
Median Age (Interquartile Range (IQR))	1.7 years (2.7)	1.2 years (2.1)	0.86 years (3.7)		1.6 years (2.1)	0.80 years (1.8)	
Male	1.6 years (2.6)	1.2 years (2.0)	1.2 years (4.3)		1.6 years (2.0)	0.71 years (1.5)	
Female	1.7 years (2.7)	1.1 years (2.2)	0.85 years (3.5)	0.60	1.6 years (2.3)	0.91 years (2.1)	0.004

[a] The 8 patients not shown in this table either showed a spontaneous reduction (n = 6) or were referred to a tertiary center (n = 2), also see Fig. 1.
[b] The 3 patients not shown in this table had primary surgical reduction after a recurrence, also see Fig. 1.
[c] chi-Square test (gender) and Mann–Whitney *U* test (median age) were used for statistical analysis. $P < 0.05$ is significant.

of data), 20 underwent surgery without a prior radiological reduction. The indications for primary surgery consisted of atypical findings on abdominal US, free abdominal fluid on US, suspected peritonitis, third recurrence of intussusception for which the patient was transferred to a pediatric surgical centre, septic presentation and intussusception thought to have been present for more than 24 h. Six showed a spontaneous reduction without intervention. There was no statistically significant difference in gender and age between patients with an ileocolic intussusception who underwent primary surgery and patients with an ileocolic intussusception who underwent radiological reduction (p = 0.28, p = 0.60).

The final study group consisted of 408 patients, 282 (69.1%) boys and 126 (30.9%) girls, who all underwent radiological reduction (Fig. 1).

2.2. Radiological reduction

All radiological reductions were performed via hydrostatic reduction. The procedure was performed either by using US or fluoroscopic guidance. All hydrostatic reductions were performed by pediatric radiologists, experienced radiologists or radiology residents under supervision. The child was admitted for surgical reduction following unsuccessful hydrostatic reduction.

2.3. Primary outcome

Our data showed that during the study period in 13 out of 112 children (11.6%) who underwent surgery after an unsuccessful hydrostatic reduction of an ileocolic intussusception, no intraoperative evidence of intussusception was found. After reviewing the radiological images of these 13 children, in 3 (2.7%) children reflux of contrast into the terminal ileum was visible; these cases are true false negative results. In 7 (6.3%) children there was filling up to the caecal valve, but no reflux of contrast into the terminal ileum was visible, we classified these cases as potential false negative results. In 3 (2.7%) children there was an intussusception visible on the radiologic images.

2.4. Secondary outcome: success rate radiologic reduction

Four hundred and eight patients with an ileocolic intussusception underwent radiological reduction (Tables 1 & 2). Of these patients, 293 were hydrostatically reduced successfully (71.8%). Children in whom hydrostatic reduction was unsuccessful were significantly younger than children who had a successful hydrostatic reduction (P = 0.004). There is no statistically significant gender difference between patients in whom hydrostatic reduction succeeded or was unsuccessful (p = 0.57).

The diagnosis of unsuccessful hydrostatic reduction was made by either a pediatric radiologist (N = 48), a radiologist with a different specialty (N = 33) or a resident in training (N = 31). In the case of the 13 children in whom no intraoperative evidence of intussusception was found, in 3 cases a pediatric radiologist, in 8 cases a radiologist with a different specialty and in 2 cases a resident in training confirmed the diagnosis.

2.5. Secondary outcome: surgical reduction

After an unsuccessful hydrostatic reduction, 112 children were admitted for surgical reduction. At the discretion of the surgeon either a laparotomy or laparoscopy was initiated.

Prior to closure, the bowel was carefully inspected for evidence of ischemia, perforation and the presence of a lead point. If a lead point was present or partial resection of the bowel was conducted, specimens were obtained for histopathological analysis.

Of the surgical patients 91 (81.3%) were operated by laparotomy and 21 (18.8%) by laparoscopy. Ten (47.6%) of the 21 patients treated by laparoscopy were converted to laparotomy.

2.6. Secondary outcome: lead point distribution

Table 3 shows the distribution of lead points between non-surgically treated patients and surgically treated patients. The cause of intussusception in the 293 non-surgically treated patients was determined based on US findings.

Of the 112 surgically treated patients, histopathological analysis of surgical specimens was available in 67 cases. This confirmed a pathologic lead point in 41 (61.2%) patients. The cause of intussusception in the 45 surgically treated patients who did not have material suitable for histopathological analysis, was determined by sonography or by observations made during the operation. In 38 of these 45 patients a lead point was shown (84.4%) (Table 4).

3. Discussion

The present study is a retrospective study of a cohort of 593 patients with a suspected intussusception of whom 408 children with an ileocolic intussusception underwent hydrostatic reduction. Of these patients, 112 underwent surgical exploration after an unsuccessful hydrostatic reduction. This study shows that 11.6% of all children (13/112) who underwent surgery after an unsuccessful hydrostatic reduction of an ileocolic intussusception had no intraoperative evidence of intussusception.

This is the first study to specifically examine the incidence of negative surgical exploration after unsuccessful hydrostatic reduction of ileocolic intussusception in children. Literature related to operative findings after unsuccessful hydrostatic reduction of an ileocolic intussusception is scarce. Kia et al. mentioned that approximately 10% of all intussusceptions failing radiologic reduction, will have spontaneously reduced by the time of surgery [10]. This percentage is, however,

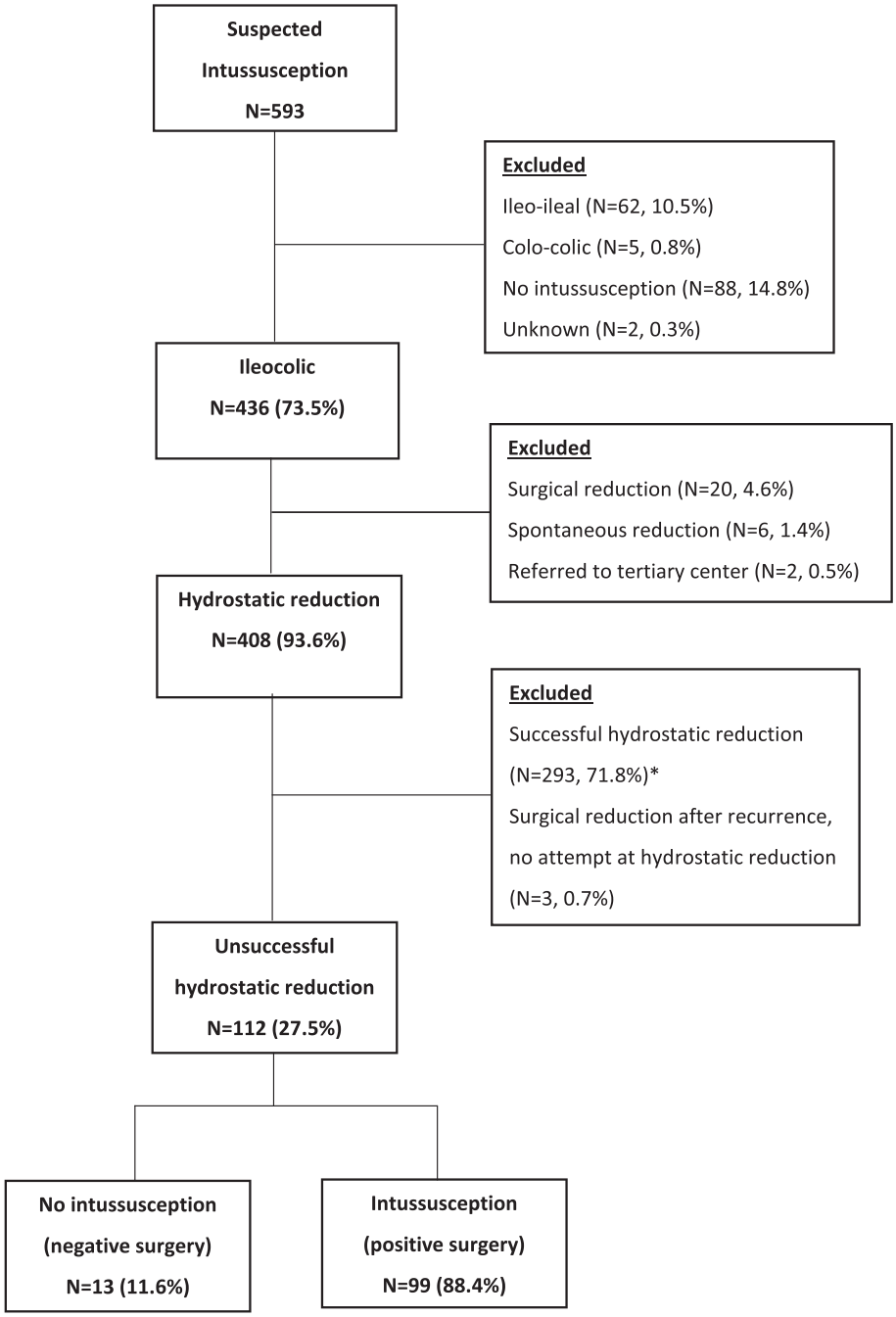

Fig. 1. Flow of participants through the study according to the STARD guideline [32]. *One patient had an unsuccessful hydrostatic reduction but at another attempt 1 day later seemed to have no intussusception anymore. This patient underwent surgical intervention because of pathologic findings at ultrasound examination.

based on the results of other studies conducted more than 25 years ago (1971–1989). In a more recent study, Naiditch et al. evaluated the operative findings after an unsuccessful primary enema for intussusception [11]. They found that of the 38 patients who underwent operative exploration after a failed primary enema, 4 patients (10.5%) had a negative intraoperative finding. However, this study described a small study population (N = 38) and they did not describe this subpopulation in detail. Ntoulia et al. evaluated the correlation between radiologic, surgical and pathologic findings in children with a failed reduction of the intussusception [12]. They found that of 72 children who underwent surgery after failed reduction, 5 children (6.9%) had no intraoperative evidence of intussusception. However, this result was not the main focus of their study and they did not draw conclusions based on this outcome. In a study by Pierro et al. the authors showed that in 11.4% of their patients reflux of contrast into the terminal ileum following apparently complete reduction of an intussusception by contrast enema was absent [13]. In none of these cases additional treatment was performed. In their surgical group they found no negative surgeries. This could explain the fact that after reviewing the radiologic images of the children who had negative intraoperative findings at laparoscopy or laparotomy in our study, 7 children showing no reflux of contrast in the terminal ileum, eventually had negative intraoperative findings at surgery.

All patients in our study with an ileocolic intussusception, underwent hydrostatic reduction [14,15]. The success percentage of hydrostatic reduction in comparable studies ranges between 69.6–82.6% [12,14,15]. The success percentage of hydrostatic reduction in our study is 71.8%, this is consistent with known literature.

Table 2
Overview of reduction protocols.

	Fluid column		Fluid type	Sedation
	Height above table	Pressure*		
Academic Medical Centre	100–120 cm	73.82–88.59 mmHg	Telebrix 1:1 water	None
Vrije Universiteit Medical Centre	50 cm	36.91 mmHg	Telebrix, gastro- or urografin	None
University Medical Centre Utrecht	150 cm	110.73 mmHg	Urografin 1:3 tapwater	Yes, eventually after third try within 1 attempt
University Medical Centre Groningen	Highest position IV pole	Depending on height IV pole	Telebrix 12S	None
Erasmus Medical Centre	150 cm	110.73 mmHg	Urografin 30% 1:1 water	None
Radboud University Medical Centre	100 cm	73.82 mmHg	Telebrix 12S 1:1 water	None
Maastricht University Medical Centre +	100 cm 150 cm after second try within one attempt	73.82 mmHg 110.73 mmHg	Telebrix 12S 1:1 water	None
Juliana Children's Hospital	100 cm 120 cm after second try within one attempt	73.82 mmHg 88.59 mmHg	Omnipaque 140 4:6 diluted	Yes, eventually after second try within one attempt (IV valium)
Albert Schweitzer Hospital	100 cm 120 cm after first try within one attempt	73.82 mmHg, 88.59 mmHg	Omnipaque 140 1:2 diluted	Yes, eventually after second try within one attempt (IV valium)

Formula: $p = h*g*rho$
$h_{water}*g*rho_{water} = P_0 = h_{Hg}*g*rho_{Hg}$ (g = equal)
$h_{water}*rho_{water} = h_{Hg}*rho_{Hg}$
h_{water} [mmH2O] = h_{Hg} [mmHg]*rho_{Hg}/rho_{water}
$mmH_2O = mmHg*13.546$
$mmHg = mmH_20/13.546$
Example:
30–168 cmH_20 = 22–124 mmHg
80–120 mmHg = 108–163 cm

p = pressure.
h = height.
g = gravity.
rho = liquid density.
* Formula conversion liquid pressure (mmH2O) to air pressure (mmHg).

Surgical reduction was performed in 112 children after an unsuccessful hydrostatic reduction. Usually a laparotomy is performed because the technique in reducing the intussusception consists of manual compression on the bowel forcing the head of the intussusception in a proximal direction. Traction on the bowel, as applied during laparoscopy, has long been controversial as it has been suggested that it could lead to perforation of the bowel [18]. However, several studies have indicated that laparoscopy might be a safe and effective method of reducing intussusception in certain patient populations, with conversion rates ranging between 12.5–31.9% [10,16–19]. In our study, 21 children underwent primary laparoscopy after unsuccessful hydrostatic reduction (18.8%) with a conversion rate of 47.6%. Laparoscopic surgery is not a standardized treatment for intussusception in the Netherlands. This could explain both the relative low number of patients that underwent laparoscopy as well as the relative high conversion rate in our study.

Most intussusceptions are not caused by a pathologic lesion and are therefore called idiopathic intussusception. The word idiopathic has also been used in literature to describe intussusceptions caused by enlarged lymph nodes, lymphoid hyperplasia or Peyer's patches because these are variations in normal anatomy as a reaction on an inflammatory process. These studies define pathologic lesions at the lead point of intussusception other than enlarged lymph nodes, lymphoid hyperplasia or Peyer's patches as pathologic lead points [20–23]. Pathologic lead points can be identified in about 6% (range 1.5–12%) of all episodes of intussusception [23–25]. The most common cause is a Meckel diverticulum, followed by duplication cysts, polyps and lymphoma [20,23]. Our results showed that 5.6% of the ileocolic intussusceptions in our study were caused by evident pathologic lead points. (Tables 2 & 3).

The strength of our study is that this, to our knowledge, is the first study in literature focusing specifically on determining the percentage of children have a negative intraoperative findings after an unsuccessful hydrostatic reduction. The study population is large enough to draw conclusions. And as the incidence of intussusception and successful radiological reduction rate in the Netherlands is comparable to other western countries, we feel that our results are representative for a larger audience.

Table 3
Cause of the intussusception (N = 408).

	Non-surgical (N = 293)	Surgical PA* (N = 69)	Surgical no PA* (N = 46)	Total (%)
Enlarged lymph nodes/mesenteric lymphadenitis	171	8	26	205 (50.2%)
Peyer's patch	4	8	10	22 (5.4%)
Lymphoid hyperplasia	3	10	0	13 (3.2%)
Meckel diverticulum	1	8	0	9 (2.2%)
Duplication cyst	0	4	0	4 (1.0%)
Cyst (non-duplication)	0	1	0	1 (0.2%)
Henoch-Schönlein purpura	0	1	2	3 (0.7%)
Burkitt lymphoma	0	2	0	2 (0.5%)
Polyp	1	0	0	1 (0.2%)
Appendicitis	0	2	0	2 (0.5%)
Remnant omphalomesenteric duct	0	1	0	1 (0.2%)
No leadpoint/non-specific findings	113	24	8	145 (35.5%)

* PA = histopathological analysis.

Table 4
Cause of the intussusception in the surgical patient group (N = 112).

	PA*	No PA*	Total (%)
Enlarged lymph nodes/mesenteric lymphadenitis	7	26	33 (29.5%)
Peyer's patch	7	10	17 (15.2%)
Lymphoid hyperplasia	9	0	9 (8.0%)
Meckel diverticulum	8	0	8 (7.1%)
Duplication cyst	4	0	4 (3.6%)
Cyst (non-duplication)	1	0	1 (0.9%)
Henoch-Schönlein purpura	0	2	2 (1.8%)
Burkitt lymphoma	2	0	2 (1.8%)
Polyp	0	0	0 (0.0%)
Appendicitis	2	0	2 (1.8%)
Remnant omphalomesenteric duct	1	0	1 (0.9%)
No leadpoint/non-specific findings	26	7	33 (29.5%)

* PA = histopathological analysis.

There are also limitations to our study, the main limitation is related to its retrospective design. There could be selection bias in the patients undergoing surgery after an unsuccessful reduction as a result of the different radiological protocols used among the participating centres in this study. The risk of the use of different protocols is the possibility of a non-optimal primary reduction, yielding surgery to a patient in one centre, whilst a slightly different approach leads to a successful reduction in another centre.

Another limitation is the possibility that some of the reductions were performed by less experienced radiologists, yielding an unsuccessful reduction when, in the hands of an experienced radiologist, it would have been a successful reduction. As shown there were false negative diagnoses on fluoroscopy. In some cases, it was difficult to distinguish an oedematous valve of Bauhin from an incomplete reduction. We could only rely on static images showing a possible persistent intussusception after an attempt of hydrostatic reduction. It would have been better to have the advantage of real-time evaluation by an experienced pediatric radiologist. We hypothesize that the presence of an oedematous valve of Bauhin could be the cause of the absence of reflux of contrast in the terminal ileum even if an intussusception actually has been reduced.

Another limitation is the fact that during the period studied there was no routine protocol for delayed reduction, i.e. it was left to the discretion of the treating team to attempt a delayed procedure. Due to the retrospective nature of the study it was not possible to analyze the number of attempts or sessions, which is therefore not included in this studies. Studies have shown that a delayed attempt can achieve a significant proportion of successful reductions, in the range of 39% to 70% [11,26–31]. We therefore believe that if delayed attempts would have been part of standard care the number of negative intraoperative findings would have been lower. The current results added to the nationwide guideline on the treatment of intussusception in the Netherlands with clear recommendations on repeat attempts.

A final limitation is the diagnosis of the cause of the intussusception in the non-surgical group. In only a minority of cases 69 out of 408 cases (16.9%) a histopathological diagnosis of the cause of the intussusception was available. Therefore we based the cause in the majority of cases on the US findings as reported by the radiologist performing the exam or on the visual inspection findings as reported by the surgeon.

Our study shows that a substantial number of children had no intraoperative finding of intussusception after an unsuccessful hydrostatic reduction. Considering this finding, we feel that this potential outcome should be included in the informed consent of the surgical procedure after an unsuccessful hydrostatic reduction. When surgery is necessary we suggest initiating laparoscopy instead of laparotomy to further decrease (post-)operative morbidity.

4. Conclusion

The results of this study show that a substantial number of children (11.6%) undergo surgery after unsuccessful hydrostatic reduction in whom no ileocolic intussusception is found intraoperatively. This could be explained by diagnostic error of radiology and ultrasound, effect of anesthesia in relaxing smooth muscle and possible effects of mechanical manipulation during surgical exposure. We would like to emphasize that spontaneous reduction is not a 'risk', but the safest of all possible sequelae. We suggest addressing this possible outcome in the informed consent of the surgical exploration. Furthermore, we suggest initiating laparoscopy instead of laparotomy when surgery is necessary.

Acknowledgements/ Collaborators

The following are members of the Dutch intussusception group: J.H. Allema (Department of Pediatric Surgery, Juliana Children's Hospital – HAGA 'Teaching Hospital', the Hague), J.L.M. Bruggink (Department of Pediatric Surgery, University Medical Centre Groningen), M.Y. van Herwaarden (Department of Pediatric Surgery, University Medical Centre Utrecht), H.C. Holscher (Department of Pediatric Radiology, Juliana Children's Hospital - HAGA 'Teaching Hospital', the Hague), W.M. Klein (Department of Radiology and Nuclear Medicine, Radboud University Medical Centre, Nijmegen) R.A.J. Nievelstein (Department of Radiology, University Medical Centre Utrecht), S.G.F. Robben (Department of Radiology, Maastricht University Medical Centre +), J. van Schuppen (Department of Radiology, University Medical Centre Groningen), J.I.L.M. Verbeke (Department of Radiology, VU University Medical Centre, Amsterdam), R. Wijnen (Department of Pediatric Surgery, Sophia Children's Hospital, Erasmus University Medical Centre).

References

[1] Buettcher M, Baer G, Bonhoeffer J, et al. Three-year surveillance of intussusception in children in Switzerland. Pediatrics 2007;120(3):473–80.
[2] Harrington L, Connolly B, Hu X, et al. Ultrasonographic and clinical predictors of intussusception. J Pediatr 1998;132(5):836–9.
[3] Cera SM. Intestinal intussusception. Clin Colon Rectal Surg 2008;21(2):106–13.
[4] West KW, Stephens B, Vane DW, et al. Intussusception: Current management in infants and children. Surgery 1987;102(4):704–10.
[5] Mandeville K, Chien M, Willyerd FA, et al. Intussusception: clinical presentations and imaging characteristics. Pediatr Emerg Care 2012;28(9):842–4.
[6] Rubinstein JC, Liu L, Caty MG, et al. Pathologic leadpoint is uncommon in ileo-colic intussusception regardless of age. J Pediatr Surg 2015;50(10):1665–7.
[7] Bines JE, IB. Acute intussusception in infants and children: a global perspective. A report prepared for the steering committee on Diarrhoeal disease vaccines, vaccine Development,Vaccines and biologicals. Vaccines and biologicalsGeneva, Switzerland: World Health Organization; 2002 [02.19].
[8] The Statistics Netherlands (SN). The electronic data recording system. [9th June 2016]. Available from: http://statline.cbs.nl/StatWeb/publication/?DM=SLNL&PA=7461BEV&D1=0&D2=1-2&D3=0-100&D4=0,10,20,30,40,50,l&HDR=T,G3&STB=G1,G2&VW=T.
[9] The Dutch Health Authority. Open data diagnosis related group (DRG) registration [19th June 2016]. Available from: http://www.opendisdata.nl/.
[10] Kia KF, Mony VK, Drongowski RA, et al. Laparoscopic vs open surgical approach for intussusception requiring operative intervention. J Pediatr Surg 2005;40(1):281–4.
[11] Naiditch JA, Rigsby C, Chin A. Delayed repeated enema and operative findings after unsuccessful primary enema for intussusception. Eur J Pediatr Surg 2012;22(5):404–8.
[12] Ntoulia A, Tharakan SJ, Reid JR, et al. Failed intussusception reduction in children: correlation between radiologic, surgical, and pathologic findings. AJR Am J Roentgenol 2016:1–10.
[13] Pierro A, Donnell SC, Paraskevopoulou C, et al. Indications for laparotomy after hydrostatic reduction for intussusception. J Pediatr Surg 1993;28(9):1154–7.
[14] Sadigh G, Zou KH, Razavi SA, et al. Meta-analysis of air versus liquid enema for intussusception reduction in children. AJR Am J Roentgenol 2015;205(5):W542–9.
[15] Beres AL, Baird R. An institutional analysis and systematic review with meta-analysis of pneumatic versus hydrostatic reduction for pediatric intussusception. Surgery 2013;154(2):328–34.
[16] van der Laan M, Bax NM, van der Zee DC, et al. The role of laparoscopy in the management of childhood intussusception. Surg Endosc 2001;15(4):373–6.
[17] Poddoubnyi IV, Dronov AF, Blinnikov OI, et al. Laparoscopy in the treatment of intussusception in children. J Pediatr Surg 1998;33(8):1194–7.
[18] Bailey KA, Wales PW, Gerstle JT. Laparoscopic versus open reduction of intussusception in children: a single-institution comparative experience. J Pediatr Surg 2007;42(5):845–8.
[19] Bonnard A, Demarche M, Dimitriu C, et al. Indications for laparoscopy in the management of intussusception: a multicentre retrospective study conducted by the French study Group for Pediatric Laparoscopy (GECI). J Pediatr Surg 2008;43(7):1249–53.
[20] Navarro O, Dugougeat F, Kornecki A, et al. The impact of imaging in the management of intussusception owing to pathologic lead points in children. A review of 43 cases. Pediatr Radiol 2000;30(9):594–603.

[21] Daneman A, Navarro O. Intussusception. Part 1: a review of diagnostic approaches. Pediatr Radiol 2003;33(2):79–85.
[22] Daneman A, Navarro O. Intussusception. Part 2: an update on the evolution of management. Pediatr Radiol 2004;34(2):97–108 [quiz 87].
[23] Applegate KE. Intussusception in children: evidence-based diagnosis and treatment. Pediatr Radiol 2009;39(Suppl. 2):S140–3.
[24] Blakelock RT, Beasley SW. The clinical implications of non-idiopathic intussusception. Pediatr Surg Int 1998;14(3):163–7.
[25] Navarro O, Daneman A. Intussusception. Part 3: diagnosis and management of those with an identifiable or predisposing cause and those that reduce spontaneously. Pediatr Radiol 2004;34(4):305–12 [quiz 69].
[26] Curtis JL, Gutierrez IM, Kirk SR, et al. Failure of enema reduction for ileocolic intussusception at a referring hospital does not preclude repeat attempts at a children's hospital. J Pediatr Surg 2010;45(6):1178–81.
[27] Gorenstein A, Raucher A, Serour F, et al. Intussusception in children: reduction with repeated, delayed air enema. Radiology 1998;206(3):721–4.
[28] Lautz TB, Thurm CW, Rothstein DH. Delayed repeat enemas are safe and cost effective in the management of pediatric intussusception. J Pediatr Surg 2015;50(3):423–7.
[29] Navarro OM, Daneman A, Chae A. Intussusception: The use of delayed, repeated reduction attempts and the management of intussusceptions due to pathologic lead points in pediatric patients. AJR Am J Roentgenol 2004;182(5):1169–76.
[30] Pazo A, Hill J, Losek JD. Delayed repeat enema in the management of intussusception. Pediatr Emerg Care 2010;26(9):640–5.
[31] Saxton V, Katz M, Phelan E, et al. Intussusception: a repeat delayed gas enema increases the nonoperative reduction rate. J Pediatr Surg 1994;29(5):588–9.
[32] Bossuyt PM, Reitsma JB, Bruns DE, et al. STARD 2015: an updated list of essential items for reporting diagnostic accuracy studies. BMJ 2015;h5527:351.