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Meraj N. Ondhia, Yousef Al-Mutawa, Srikrishna Harave, Paul D. Losty

PII: S0022-3468(19)30555-X

DOI: https://doi.org/10.1016/j.jpedsurg.2019.07.022

Reference: YJPSU 59307

To appear in: *Journal of Pediatric Surgery*

Received date: 27 May 2019

Revised date: 24 July 2019

Accepted date: 25 July 2019

Please cite this article as: M.N. Ondhia, Y. Al-Mutawa, S. Harave, et al., Intussusception: a 14-year experience at a UK tertiary referral Centre, *Journal of Pediatric Surgery*(2019), https://doi.org/10.1016/j.jpedsurg.2019.07.022

Intussusception: A 14-Year Experience At A UK Tertiary Referral Centre

Meraj N. Ondhia[1], Yousef Al-Mutawa[1], Srikrishna Harave[2], Paul D. Losty[1,3]

[1]Institute of Child Health, University of Liverpool, Liverpool, United Kingdom.

[2]Department of Paediatric Radiology, Alder Hey Children's Hospital NHS Foundation Trust, Liverpool, United Kingdom

[3]Department of Paediatric Surgery, Alder Hey Children's Hospital NHS Foundation Trust, Liverpool, United Kingdom

Meraj Ondhia and Yousef Al-Mutawa contributed as co-authors

Corresponding author: Paul D Losty

Corresponding author email: paul.losty@liverpool.ac.uk

Corresponding author postal address: Department of Paediatric Surgery, Alder Hey Children's Hospital NHS Foundation Trust, East Prescot Rd, Liverpool, L14 5AB, UK

Abstract

Aims

Intussusception is the most common cause of acute intestinal obstruction in infants. First line management in uncomplicated cases at our centre is fluoroscopy guided air enema reduction. This study reports a 14 year UK single centre experience highlighting management and outcomes of intussusception in children.

Methods

All cases of intussusception (ICD 10-code K56.1) at a single tertiary referral centre from 2004 to 2017 were analysed. Data evaluated included patient demographics, clinical presentation, treatment modality(s), complications and outcome(s).

Results

290 confirmed cases (69% male) of intussusception were identified during the study period. The median age at presentation was 9.5 months (range 4 days – 15 years). 186 (64%) cases occurred in children who were transferred to Alder Hey from peripheral district hospitals. 196 cases (68%) proceeded directly to air enema reduction and successful reduction was achieved in 129 cases (66%). A single case (0.5%) of attempted air enema reduction was complicated by perforation. Early recurrence rate following air enema reduction was 9%. Operative management was indicated in 140 (48%) cases of which 66 required bowel resection (47%). There were no deaths.

Conclusions

We highlight one of the largest single centre UK studies defining practice outcomes for intussusception across the British Isles. Our centre has achieved a 66% success rate of non-operative reduction over 14 years in line with the British Society Paediatric Radiology (BSPR) recommendations and within 5% of the national median metric (71%). Moreover these outcomes were achieved with a very low complication rate (0.5%) from air enema reduction and no mortality in the series.

Type of study

Retrospective study

Level of evidence

III

Keywords

Intussusception, fluoroscopy-guided air enema reduction

Intussusception is the invagination of a portion of proximal bowel (intussusceptum) into the adjacent distal bowel (intussuscipiens) with a peak incidence at 6-18 months of age [1]. The aetiology is idiopathic in 90% of cases [2] and the lead point is frequently attributed to hypertrophic lymphoid tissue(s) within the gut wall following presumed infection(s) – viral or bacterial [3]. The frequently described triad of abdominal pain, redcurrant jelly stool, and presence of an abdominal mass is not regularly detected in the clinical setting, young infants and toddlers more often presenting with a gastroenteritis-like illness [4]. Prompt diagnosis based on clinical suspicion should be confirmed with abdominal ultrasonography, which has shown almost 100% sensitivity and specificity of 80-100%[5].

The widely accepted first-line treatment modality is non-operative reduction, performed at our institution by paediatric radiologists using fluoroscopy guided air enema. Surgical intervention is undertaken in cases of perforation, bowel necrosis and/or unsuccessful non-operative reduction. From the literature it is evident there is wide variation in the reported success rate(s) of non-operative reduction between individual centres (38 – 90%) and the number of index cases performed per centre (0 – 31/year) [6, 7]. Against this background this study therefore reports the outcomes of children with intussusception presenting to our paediatric surgical centre over a 14-year period, with particular focus on the success rate of non-operative reduction.

1. Methods

Following approval and registration as a clinical audit, case records of all patients admitted to Alder Hey Children's Hospital (a major UK tertiary referral centre) for management of intussusception (K56.1 ICD-10) for the 14-year period between January 2004 to December 2017 were examined. All cases either presented directly to our institution or were transferred from referring peripheral hospitals for definitive care. Following clinical assessment and fluid resuscitation by the paediatric surgical team, the radiology service was consulted for ultrasound scans in patients to confirm the diagnosis. Once the diagnosis was confirmed, a decision was made whether to attempt pneumatic reduction or perform surgery. First line emergent management at our institution is to schedule pneumatic reduction under fluoroscopic guidance. Each patient was carefully evaluated and considered for pneumatic reduction if haemodynamically stable. Indications for 'up front' surgery without attempted air enema reduction were: hypovolaemic shock, peritonitis or pneumoperitoneum. Unfavourable ultrasound features, including reduced or absent blood flow in the intussusception on Doppler evaluation and the presence of trapped fluid between the loops of intussusception, were considered as a relative contraindication to attempting pneumatic reduction, as they are known to affect the success rate [8].

Pneumatic reduction with fluoroscopic guidance was performed by paediatric radiologists, with a paediatric surgeon resident and/or consultant surgeon present. Intravenous morphine analgesia was carefully administered by surgeons where necessary dependent on the clinical condition of the child, as assessed by the care team prior to the radiology enema procedure For pneumatic reduction, air is insufflated via a Foley catheter (12-Fr to 16-Fr, depending on the patient's size) placed inside the patient's rectum. To achieve reduction lower air pressures are initially utilised, up to 80 mmHg, and increased to a maximum of 120 mmHg. Progress of the intussusception reduction is monitored with fluoroscopy screening at periodic intervals, to minimise the radiation dose. Our radiologists will

perform a maximum of three attempts. Successful reduction is demonstrated by free flow of air into the terminal ileum and disappearance of the soft tissue mass of the intussusception. The success rate(s) of air enema was defined as successful reductions as a proportion of all air enema performed. For patients who required operative reduction, the intussusception was released carefully by manual compression or the affected segment of bowel was resected for cases complicated by bowel necrosis. All patients who underwent operative reduction were reviewed in the paediatric surgery outpatient clinic. Early recurrence of intussusception was defined as those occurring within 72 hours of a resolved episode.

For each patient the following variables were recorded and analysed: (1) patient demographics, (2) clinical presentation, (3) imaging modality, (3) management, (4) complications.

Results are expressed as counts (n=) and percentages. Continuous variables are presented as median with range or mean ± standard deviation. An independent student's T-test was applied for continuous variables. Categorical variables were compared with Chi squared and Fisher exact test. Results were deemed to reach significance at $p < 0.05$ (2-tailed). The statistical package SPSS 24.0 (SPSS, Chicago, IL) was used for all analysis.

2. Results

290 cases of intussusception were managed in 265 patients during the study period (Figure 1). 186 (64%) cases occurred in children who were transferred to Alder Hey from peripheral hospitals. The median number of cases managed per year was 21 (range, 10 - 34). 200 (69%) episodes occurred in males. The median age at presentation was 9.5 months (range, 4 days to 15 years) and hospital stay 3 days (range, 0 to 16 days). The most common presenting clinical features were abdominal pain, n=175 (60%), vomiting, n=173 (60%) and rectal bleeding/redcurrant jelly stool, n=105(36%) cases. The classic triad of abdominal pain, vomiting and rectal bleeding was encountered in only 48/290 cases (17%). The mean duration of symptoms was 3 (±6.4) days. Ultrasonography was the primary imaging modality employed in 283 (97.6%) cases.

Air enema reduction was undertaken as a primary intervention in 196 (68%) cases. Successful reduction was achieved in 129 cases (66%). Table 1 details the success rate of air enema by year. Pressures used for air enema reduction were detailed for 133 (68%) attempted air enema reductions and the median pressure deployed was 100 mmHg (range, 50 – 125). The number of attempted reductions was detailed for 143 (73%) air enema reductions and the median number of attempts was 2 (range, 1 – 3). Only 1 single (0.5%) attempted air enema reduction was complicated by perforation. This event was in an 8 month old infant and occurred at a pressure of 110 mmHg. At surgery a perforation in an ischaemic area of the colon was found requiring a limited right-hemicolectomy. A single, n=1 (0.5%) attempted air enema reduction was abandoned in a patient due to suspicion of bowel perforation; however perforation was not detected at laparotomy.

We analysed risk factors for unsuccessful air enema reduction. The presence of a pathological lead point significantly increased the risk of air enema being unsuccessful (risk ratio=3.4, $p<0.001$). Air enema reduction was also less likely to be successful in patients transferred from a peripheral district hospital (risk ratio=1.8, $p=0.019$). The duration of symptoms, sex, age and the presence of rectal bleeding did not affect the risk of air enema failure.Patients in whom successful reduction of

the intussusception was obtained by air enema had a significantly shorter hospital stay compared with patients who required surgery as definitive management, 2.4 (± 1.8) vs. 5.2 (± 2.9) days (p<0.001).

Operative reduction was undertaken as a primary intervention without attempted air enema reduction in 73 (25%) cases and following unsuccessful air enema reduction in 67 (23%) cases. Operative reduction with bowel resection was undertaken in 66 (23%) cases, of which 51 (77%) and 15 (23%) were small and large bowel resections, respectively. Manual reduction without bowel resection was performed in 72 (25%) cases.In 2 patients the intussusception was found to have spontaneously reduced. Table 2 details the procedures performed.Of 140 patients who required operative intervention, 103 (74%) were transferred from a peripheral district hospitals. Compared with presenting directly to our hospital, being transferred from a peripheral district hospital conferred a greater risk of a patient requiring surgery as definitive management of intussusception (risk ratio=1.7, p<0.0001).

Two post-operative complications following operative reduction were encountered. A single infant child continued to vomit following open reduction and required a second look laparotomy with release of intra-abdominal adhesions. Another patient who had open reduction developed a post-operative bowel perforation requiring a right hemicolectomy.The risk(s) of a child requiring bowel resection were significantly increased when surgery was undertaken as the primary intervention compared with those patients who had unsuccessful air enema reduction (risk ratio=1.7, p=0.004).

In 21 (7%) cases intussusception was diagnosed by ultrasound but a therapeutic intervention was not required due to spontaneous resolution of the intussusception on further imaging given the transient nature of the intussusception.

In 54 (17%) cases the intussusception was associated with a pathological lead point (PLP) (Table 3). Intussusception was the initial primary presentation of a diffuse large B-cell and Burkitt's lymphoma in 3 (1%) and 2 (0.7%) cases, respectively. The mean age of children with intussusception with a PLP compared with idiopathic intussusception cases was 60 (± 15.4) vs. 14.9 (± 19.0) months (p<0.001).

Overall, 25 confirmed recurrences of intussusception were encountered during the study period. Nineteen patients had a single recurrence and 3 patients had 2 confirmed recurrences of intussusception. There were 12 early recurrences (within 72 hours), all of which were following successful air enema, giving an early recurrence rate following air enema reduction of 9%. 8 (67%) early recurrences following air enema reduction were successfully reduced by repeat air enema and 4 (33%) cases required operative reduction. 13 recurrences occurred more than 72 hours after the initial episode and the median time to recurrence was 121 days (range, 14 – 2184).

3. Discussion

Intussusception is a paediatric surgical emergency which requires timely diagnosis and management. The majority of patients can avoid an operation and fully recover when managed by skilled paediatric radiologists and paediatric surgeons.

Consistent with published studies, in this single centre study, 219/290 (75%) of cases occurred in patients under 2 years and 200/290 (69%) of the index cases occurred in males (1.5:1)[1]. Air enema

reduction was performed as a first line intervention in 196/290 (68%) cases managed at our centre. Outcome metrics used to benchmark standards and quality of care in managing childhood intussusception are integrally linked with the success rate of air enema reduction, mitigating the need for surgical intervention. The success rate achieved at our UK centre with air enema reduction was 66% during a 14 year study period. Samad et al. have previously reported a success rate of 61% for air enema reduction across the UK and Ireland [6]. A more recent national-level surveillance study conducted by Hannon et al. reported that intussusception reduction rates varied widely (31-90%) across individual centres with a national reduction rate/target figure quoted by this audit at 71% success [7]. Our data equate well with previous UK national audit studies which have made efforts to 'benchmark' nationwide reduction rates along with variations in practice across paediatric hospitals [6, 7]. Non-operative reduction has a high overall success rate and low complication and recurrence rates. It also leads to a shorter hospital stay and is therefore recommended as the first-line treatment of this condition where possible.

Intestinal perforation is a recognised complication of air enema reduction. The aetiology of perforation during attempted air enema reduction is multi-factorial including bowel wall ischaemia, patient and technical factors [9]. Perforation rate(s) are widely regarded as a marker of performance and safety and a recent comprehensive review suggests contemporary perforation rates internationally are generally low (<1%) for pneumatic reduction [10]. At our UK centre the perforation rate for air enema reduction was only 0.5%, this compares favourably with international data and the most recent UK national audit which found a perforation rate of 2.5% [7].

A number of studies have suggested that children referred to specialist centres for definitive management of intussusception have a higher rate of surgical intervention [11, 12]. Our results were consistent with these published reports and we found patients who were transferred were more likely to require surgery as definitive management of intussusception. In addition, air enema reduction was significantly less likely to be successful in paediatric patients transferred from district regional hospitals to our centre. Here it is inferred with interhospital transfer patients experienced delay in having definitive management of intussusception catered for at a specialist paediatric surgery centre.

In this study a pathological lead point was identified in 54 (19%) patients. Meckel's diverticulum (n=15), Henoch Schonlein Purpura (n=15), lymphoma (n=5), hamartomatous polyps (n=3) are all established entities in childhood intussusception [13]. Lymphoid hyperplasia was identified on histopathology examination of resected bowel specimens in 2.3% of cases. Lymphoid hyperplasia secondary to viral or other infections is well recognised in the likely aetiology and pathogenesis of most index cases of childhood idiopathic intussusception [14, 15].

It is noteworthy there is relatively sparse literature discussing the recurrence of intussusception following both operative and non-operative reduction of intussusception. A recent meta-analysis by Gray et al. reported early recurrence rates following fluoroscopy guided air enema were 6.9-10.4% [16]. In this series we observed an early recurrence rate of 9% following air enema reduction, in line with results from this meta-analysis. No early recurrences were observed following operative reduction.

We acknowledge the limitations of this study owing to the retrospective nature of the data collection. We utilised medical coding (K56.1 ICD-10) at our hospital to identify patients with intussusception and the quality of data collected was dependent on robust medical records.

In conclusion, we highlight one of the largest single centre UK studies defining practice outcomes for intussusception across the British Isles. Success rates with non operative air enema reduction at Alder Hey Children's Hospital compare favourably with UK nationwide metrics/ British Society of Paediatric Radiology. Finally, these outcomes were achieved with a low complication rate (0.5%) from air enema reduction and operative interventions with no deaths.

Funding
This research did not receive any specific grant(s) from funding agencies in the public, commercial, or not-for-profit sectors.

The authors declare there are no competing interests.

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Table 1: success rate of air enema by year

Year	Air enemas (n)	Success rate (%)
2004	13	62
2005	10	50
2006	8	63
2007	13	69
2008	12	50
2009	14	64
2010	20	75
2011	16	75
2012	22	68
2013	18	56
2014	12	75
2015	8	63
2016	16	75
2017	14	64

Table 2: surgical procedures undertaken

Surgical procedure (n=140)	Number of cases (%)	
	Primary	Secondary
Open reduction	22 (16%)	33 (24%)
Open reduction + appendicectomy	6 (4%)	11 (8%)
Ileal resection	27 (19%)	14 (10%)
Ileal resection + appendicectomy	8 (6%)	2 (1%)
Hemicolectomy	8 (6%)	7 (5%)
Spontaneous resolution at laparotomy	2 (1%)	0
	73	67

TABLE 3

Pathological lead point (n=54)

Pathological lead point	Number of cases (%)		Median age (range)
Meckel's diverticulum	15 (27.8)		4.9 years (3.0 years – 7.1 years)
Henoch Schonlein Purpura	15 (27.8)		6.0 years (0.1 months – 11.1 years)
Lymphoid hyperplasia	8	(14.8)	10.1 months (3.7 months – 1.6 years)
Hamartoma	3	(5.6)	10.4 years (8.5 years – 13.4 years)
Diffuse large B-cell lymphoma	3	(5.6)	9.7 years (7.8 years - 15.7 years)
Burkitt's lymphoma	2	(3.7)	4.2 years (3.5 years – 5.0 years)
Small bowel duplication cyst	2	(3.7)	2.0 months (1.7 months – 2.2 months)
Ectopic exocrine pancreatic tissue	1	(1.9)	9.4 months
Ileal lymphatic malformation	1	(1.9)	7.8 years
Inflammatory fibroid caecal polyp	1	(1.9)	9.6 years
Lymphangiectasia	1	(1.9)	1.5 years
Mesenteric lymph node	1	(1.9)	1.2 years
Pericolic lymph node	1	(1.9)	7.0 months

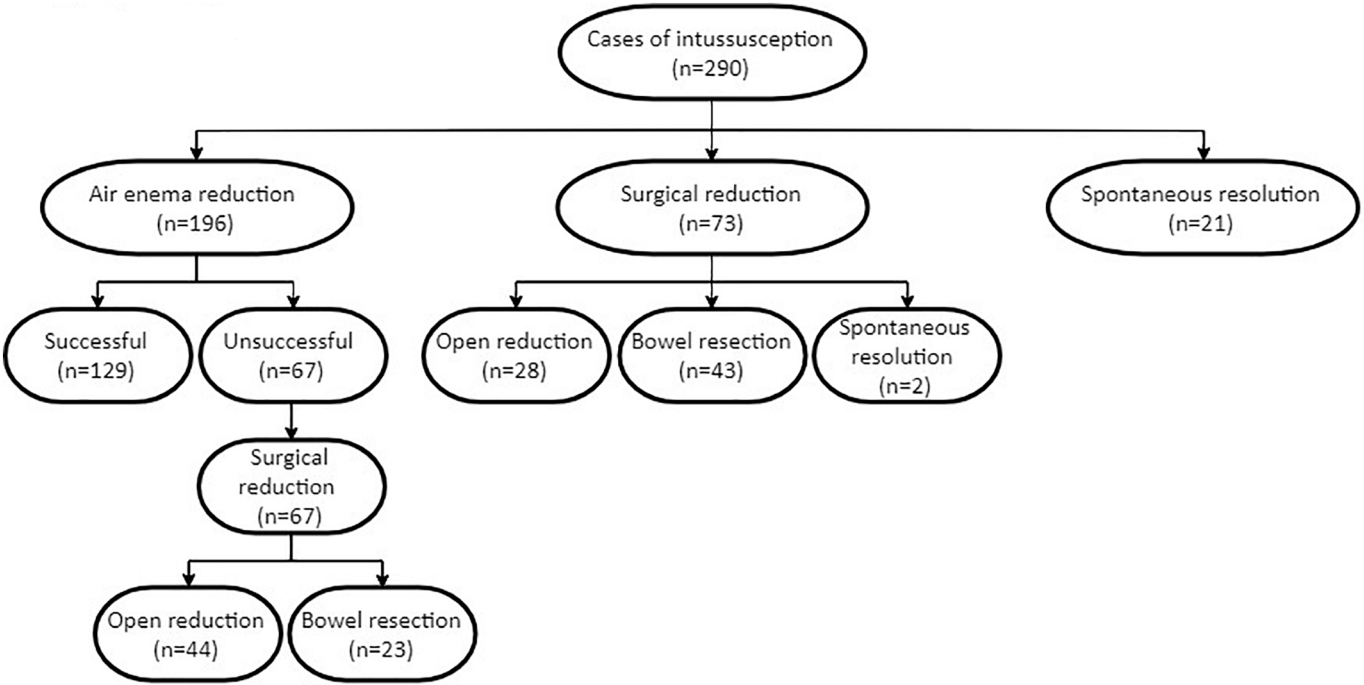
Cases of intussusception
(n=290)
Air enema reduction
(n=196)
Surgical reduction
(n=73)
Spontaneous resolution
(n=21)
Successful
(n=129)
Unsuccessful
(n=67)
Open reduction
(n=28)
Bowel resection
(n=43)
Spontaneous
resolution
(n=2)
Surgical
reduction
(n=67)
Open reduction
(n=44)
Bowel resection
(n=23)

Figure 1