Economics and clinical implications of delayed presentation of children with intussusceptions: minimizing healthcare cost in the context of limited resources

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Received 24 June 2018
Accepted 17 July 2018

The Egyptian Journal of Surgery 2018, 37:600–605

Background
A basic strategy in today's resource-limited healthcare environment is limiting cost while maintaining quality.
Objective
To estimate the impact and healthcare cost of treatment of intussusceptions (ISs) presenting late.
Patients and methods
We performed a chart review of children managed for ISs over 44 months. Our care pathway included resuscitation, ultrasound-guided hydrostatic reduction of children presenting early (≤24 h) with no peritonitis.
Results
There were 46 children seen at a median age of 8.5 months. Thirty-two (69.6%) presented late (>24 h). The total charge was higher for patients who presented late ($259.6 vs. $168.9, *P*=0.012). There was an average of 21% ($50.0) additional charges incurred for each day's delay. There was significantly higher cost of drugs ($45.1 vs. $23.7, *P*=0.008) and procedures ($154.4 vs. $99.5, *P*=0.025). Those who presented late had increased risk of bowel resection (40 vs. 0%, odds ratio=1.74, *P*=0.005), and higher complication rates (56 vs. 21%, odds ratio=0.21, *P*=0.018). The length of stay was doubled for those who presented late (10.6 vs. 4.6 days, *P*=0.001).
Conclusion
Delayed presentation of ISs translates to increased hospital cost and an extra week in hospital with increased morbidity.

Keywords:
cost, delayed presentation, hydrostatic reduction, intussusception, morbidity, ultrasound guided

Egyptian J Surgery 37:600–605

1110-1121

Introduction

Intussusception (IS) is the most common cause of acute intestinal obstruction in infants and young children. It occurs when one segment of the bowel invaginates into the adjacent distal segments, resulting in venous congestion and bowel wall edema. Consequently, obstruction of the arterial blood supply may occur leading to bowel infarction, perforation, septicemia, and even death [1]. The incidence of IS varies from 0 to 17.8 cases per 100 000 children [2] to 300 cases per 100 000 children across various regions [3–6], suggesting the differences in genetics and lifestyle factors, variations in study methods, and healthcare practices [3].

The treatment of ISs has evolved over the decades from routine operative treatment to the current trend of initial nonoperative interventional imaging pressure reduction [1,5,7,8]. Nonoperative management has been shown to shorten hospitalization and reduce morbidity and mortality [1,3,8,9]. The modern healthcare paradigm in resource-limited healthcare practice dictates limiting cost while maintaining quality.

A few studies have stressed the clinical implications of delayed presentation of IS in developing countries [8–10], but studies estimating hospital costs of ISs treatment are sparse. In order to offer an appropriate cost-effective strategy to improve the outcomes, it is very important to assess the cost of ISs management. We hypothesized that early presentation of ISs would reduce cost of care. The primary goal of our study is to estimate the healthcare cost and impact of treatment of ISs with the advent of ultrasound-guided hydrostatic reduction.

Patients and methods

We performed a retrospective chart review of children managed for ISs at a Nigerian tertiary hospital between January 2012 and August 2015. Abdominal ultrasound was used to confirm diagnosis in all the patients. Our care pathway included resuscitation, ultrasound-guided hydrostatic saline enema reduction of children presenting early (≤24 h) with no peritonitis. Patients who presented after 24 h without significant abdominal distention and peritonitis also had a trial of hydrostatic reduction but those with presence of significant abdominal distension and/or peritonitis were offered laparotomy.

Data extracted for each of the patients were their age at presentation, sex, clinical presentation, duration of symptoms before presenting to the hospital, mode of treatment, outcome of treatment, complications of surgery (complications were defined as any deviation from the normal postoperative course for an ISs surgery within 30-postoperative days), length of hospital stay, mortality, and cost of treatment. The socioeconomic status was assessed according to the Oyedeji social classification method [11]. For the purpose of this study, delayed presentation was defined as presentation after 24 h of onset of symptoms. Patients who presented within 24 h and those presenting after 24 h were compared on these variables.

Cost estimation

The direct costs incurred during hospitalization was estimated using the hospital billing records including cost of drugs, investigations, procedures, consumables, and cost of admission per patient. Indirect costs like travels, missed work hours, and missed school days, were not considered. The official exchange rate of $1=N200 was used to adjust for inflation on the cost in 2015.

The primary outcome measure was direct medical costs, and other outcome measures were postoperative complications, bowel resection, and length of stay.

Statistical analysis

Descriptive statistics were used to summarize the demographic characteristics of patients. Continuous variables were described with mean and SD or median and interquartile range. Parametric continuous variables were compared using *t*-test while nonparametric variables were compared using Mann–Whitney *U*-test. Categorical variables were described using frequencies and percentages and compared between treatment groups using χ^2-tests or Fisher's exact test. All statistical tests were two tailed, and a *P* value of less than 0.05 was considered significant. All statistical analyses were performed using SPSS for Windows 21.0 (SPSS Inc., Chicago, Illinois, USA).

Results

There were 46 children with a median age of 8.5 months (range: 3 months to 9 years). Twenty-four (52%) children were 7–12 months of age at presentation (Table 1). Seventy percent (*n*=32) were boys. Only one (2.2%) had National Health Insurance Scheme coverage. Majority (82.5%) of the children who assessed care for IS at our facility are from the upper (I and II) and middle (III) socioeconomic classes (Table 1). Fourteen (30.4%) presented early (≤24 h) and 32 (69.6%) presented late. The groups were

Table 1 Demographic characteristic of patients who presented within 24 h and those that presented after 24 h

Variables	Total (*n*=46)	Early (*n*=14)	Late (*n*=32)	*P* value
Age at diagnosis [median (range)]	8.5 (3–108)	9.0 (5–18)	8.0 (3–108)	0.728
Age group (months)				0.348
0–6	11 (23.9)	2 (14.3)	9 (28.1)	
7–12	24 (52.2)	10 (71.4)	14 (43.8)	
13–24	7 (15.2)	2 (14.3)	5 (15.6)	
>24	4 (8.7)	0 (0)	4 (12.5)	
Sex				
Male	32 (69.6)	9 (64.3)	23 (71.9)	0.607
Female	14 (30.4)	3 (21.4)	9 (28.1)	
Socioeconomic class (*n*=40)				0.874
I	5 (12.5)	1 (8.3)	4 (14.3)	
II	12 (30.0)	3 (25.0)	9 (32.1)	
III	16 (40.0)	6 (50.0)	10 (35.7)	
IV	5 (12.5)	2 (16.7)	3 (10.7)	
V	2 (5.0)	0 (0)	2 (7.1)	
Duration of symptoms (mean±SD) (h)	87.52±82.70	16.79±8.22	118.50±81.55	0.0001*

*Statistically significant.

comparable in sociodemographic characteristics as shown in Table 1.

Clinically, the most common presenting features included bilious vomiting 38 (82.6%), 'red currant jelly' stool 34 (73.9%), and abdominal mass 26 (56.5%). Other clinical presentations are shown in Table 2. Palpable abdominal mass was significantly more in those who presented early (71 vs. 50%, *P*=0.017) and abdominal distension was seen more in those who presented late (50 vs. 0%, *P*=0.008; Table 2). The triad of abdominal pain, palpable abdominal mass, and red currant jelly stools were seen in 12 (26.1%) patients.

Ultrasound guided hydrostatic saline enema reduction was 17 times more likely to be successful in those who presented early than in those who presented late [36 vs. 3%; odds ratio (OR)=17.2, *P*=0.003]. Early presentation was less likely to be associated with operation intervention (64 vs. 94%, OR=0.120, *P*=0.010). The risk of bowel resection was double in those who presented late (40 vs. 0%, OR=1.74, 95% confidence interval: 1.25–2.33, *P*=0.005), and they had a higher postoperative complication rate (56 vs. 21%, OR=0.21, 95% confidence interval: 0.043–0.80, *P*=0.018; Table 3). The postoperative complications were surgical site infections in 17 (36.9%), adhesive small bowel obstruction in three (6.5%), recurrent IS in three (6.5%), two (4.3%) each of wound dehiscence, anastomotic leak and chest infection. One (2.2%) patient among the late presenting group had iatrogenic colonic perforation. There was no significant difference in the type of postoperative

Table 2 Clinical characteristic of patients who presented within 24 h compared with those that presented after 24 h

	Total (*n*=46) [*n* (%)]	Early (*n*=14) [*n* (%)]	Late (*n*=32) [*n* (%)]	OR (confidence interval)	*P* value
Vomiting	38 (82.6)	9 (64.3)	29 (90.6)	0.466 (0.067–3.237)	0.432
Red currant stool	34 (73.9)	10 (71.4)	24 (75.0)	1.667 (0.300–9.272)	0.557
Abdominal mass	26 (56.5)	10 (71.4)	16 (50)	10.000 (1.143–87.520)	0.017*
Abdominal distension	16 (34.8)	0 (0)	16 (50)	1.533 (1.138–2.067)	0.008*
Abdominal pain/colics	15 (32.6)	4 (28.6)	11 (34.4)	0.955 (0.234–3.888)	1.000
Fever	14 (30.4)	2 (14.3)	12 (37.5)	0.333 (0.062–1.786)	0.186
Classic triad (vomiting/abdominal pain/red currant stool)	12 (26.1)	3 (21.4)	9 (28.1)	0.853 (0.187–3.883)	0.836
Anal protrusion	3 (6.5)	0 (0)	3 (9.4)	1.444 (1.161–1.721)	0.272
Loose stool	2 (4.3)	1 (7.1)	1 (3.1)	2.818 (0.162–49.008)	0.460

*Statistically significant.

Table 3 Comparison of intervention and outcomes of patients who presented within 24 h and those that presented after 24 h

	Overall (*n*=46)	Early (*n*=14)	Late (*n*=32)	OR (confidence interval)	*P* value
Successful hydrostatic reduction	6 (13.0)	5 (35.7)	1 (3.1)	17.22 (1.78–166.98)	0.003
Operative intervention	39 (84.6)	9 (64.3)	30 (93.8)	0.120 (0.02–0.73)	0.010
Bowel resection	13 (28.2)	0 (0)	13 (40.6)	1.74 (1.25–2.33)	0.005
Postoperative complications	21 (45.6)	3 (21.4)	18 (56.3)	0.212 (0.043–0.80)	0.018
SSI	17 (36.9)	3 (21.4)	14 (43.8)	0.35 (0.082–1.50)	0.149
ASBO	3 (6.5)	0 (0)	3 (9.4)	1.48 (1.21–1.83)	0.236
Recurrent	3 (6.5)	2 (14.3)	1 (3.1)	5.17 (0.43–62.40)	0.158
Wound dehiscence	2 (4.3)	1 (7.1)	1 (3.1)	2.39 (0.14–41.08)	0.539
Anastomotic leak	2 (4.3)	0 (0)	2 (6.3)	1.40 (1.16–1.70)	0.375
Chest infection	2 (4.3)	0 (0)	2 (6.3)	1.47 (1.20–1.80)	0.339
Colonic perforation[a]	1 (2.2)	0 (0)	1 (3.1)		
Death	1 (2.2)	0 (0)	1 (3.1)	0.69 (0.57–0.84)	0.504
Length of hospital stay (mean ±SD) (days)	8.76±7.42	4.57±3.55	10.59±7.96		0.001
Duration of follow-up (range) (weeks)	4 months (2 weeks to 9 months)	1 month (2 weeks to 2 years)	6 weeks (2 weeks to 2 years)		0.024

ASBO, adhesive small bowel obstruction; OR, odds ratio; SSI, surgical site infection. [a]Denote that statistical significant not done because it was an iatrogenic colonic perforation.

complications between the two groups (Table 3). The length of stay was doubled for those who presented late (10.6 vs. 4.6 days, *P*=0.001). The overall mortality was 2.2%.

Table 4 compares the direct medical cost of early and late presenters. The average total charge was higher for patients who presented late ($259.6 vs. $168.9, *P*=0.012). There was an average of 21% ($50.0) additional charges incurred for each day's delay. There was significantly higher cost of drugs ($45.1 vs. $23.7, *P*=0.008) and procedures ($154.4 vs. $99.5, *P*=0.025) among patients who presented after 24 h of onset of symptoms compared with those who presented within 24 h, respectively.

Discussion

The triad of abdominal pain, palpable abdominal mass, and red currant jelly stools has been described as the typical presentation of IS [1]. In our study, vomiting, red currant jelly stool, and palpable abdominal mass were the most prominent features at presentation, whereas abdominal pain (33%) was less common. Vomiting as a prominent feature of IS has been previously described [4,9,10]. This study has also demonstrated that the presence of red currant jelly stools was not dependent on the duration of illness, and an abdominal mass was more often palpable in patients with early (rather than late) presentation to the hospital. This was consistent with the findings in a review of 294 children with IS by Buettcher *et al.* [12], in Switzerland. The less frequent finding of abdominal pain in our study may suggest variation in the clinical presentation of IS in different regions, but it may also reflect a difference in local awareness of these symptoms by particular healthcare professionals. Abdominal pain or the perception of pain by parents was frequently reported in studies from developed countries but less often recorded in reports from developing countries [7,8,13]. This has been related to the differences in parental perceptions of what constitutes pain in children [1]. The 'classic' triad of IS (abdominal pain, red currant jelly stool, and palpable abdominal mass) is not always present in children with ISs as shown in our study. These groups of symptoms and signs are easy to recall but may lead to delayed diagnosis in patients lacking them [1].

The majority of children (69%) presented 24 h after symptom onset, which may be a hindrance to quality emergency healthcare services. This is comparable to reports from other developing countries where the majority of patients presented after 24 h of onset of symptoms [8,10,13]. However, in most developed countries most of the children were hospitalized within 24 h after symptom onset [3,12]. In a retrospective review of ISs cases in children less than 2 years of age treated during 2004–2008 at 15 hospitals in the Bukhara region of Uzbekistan, 87% presented within 24 h of symptom onset with good access to emergency healthcare services [3].

In our environment, these children were often taken to over-the-counter shops, health centers and other peripheral hospitals where they were treated for gastroenteritis and sometimes rectal prolapse, and when their clinical states deteriorated, they were then referred to tertiary hospitals for specialist management [8,14]. Poverty also contributes to the delayed presentation especially when parents seek medical care, where it is cheapest not minding the level of expertise of the healthcare provider. The limited coverage of health insurance scheme in most parts of Africa also contributed in no small measure as parents pay out of their pocket to care for their children [8].

The advent of interventional radiology has revolutionized the management of IS from operative management through ultrasound-guided hydrostatic reduction to pneumatic reduction in the developed countries [3,12]. In many developing countries, the treatment of IS is predominantly surgical. The facilities and technical expertise necessary to perform safe and effective enema reduction are frequently unavailable outside major teaching hospitals. The late presentation of a significant proportion of patients in developing countries also render them unsuitable for nonoperative reduction because of the increased risk of perforation and sepsis [1,8].

More recently, ultrasonography is not only being used for definitive diagnosis but also to guide hydrostatic reduction with saline enema in sub-Saharan Africa [8,15]. In this study, hydrostatic reduction was successful in 35.7% of patients that presented within 24 h compared with 3% in those who presented after 24 h. This finding underscores the importance and effectiveness of this technique in early presentation. This approach of IS reduction has been associated with decreased mortality and morbidity and with cost benefits associated with a reduction in the length of stay in hospital. Despite the success of enema reduction in many patients, surgery still provides an important treatment option in patients presenting with shock, complicated or recurrent IS, prolonged duration of symptoms, anal/rectal prolapse of the intussusceptum, or failed enema reduction [1].

Delay in presentation of IS increases the likelihood of surgical intervention [4,8]. Thirty-nine (84.6%) of our patients had surgical intervention slightly higher than 75% operative intervention by Ogundoyin *et al.* [8] in Ibadan, Nigeria. This is a significant improvement in developing countries when compared with previous reports where all patients had surgical intervention [7,13,16].

Latipov *et al.* [3] reported 72.7% success rate of air enema reduction in patients presented within 12 h following the onset of symptoms and 59.6% in patients treated within 24 h after symptom onset. They also noted that among children admitted within the first 24 h after the onset of illness, no intestinal resection was performed. However, four of nine children admitted more than 24 h after symptom onset had intestinal resection, with three deaths in these patients compared with a single death in children admitted earlier to the hospital [3]. In our study, bowel resection rate was 28.2%. The risk of bowel resection was double in patients who presented after 24 h (56 vs. 0%). None of those who presented within 24 h had bowel resection. Nonoperative reduction also significantly reduced the direct medical cost of treatment.

Surgical site infection was the most common postoperative complication observed in this study with higher rate in patients presenting after 24 h. Although the difference did not reach statistical significance, it significantly accounted for increased cost of postoperative wound dressing in patients presenting late. Only one patient died among those that presented after 24 h giving an overall mortality of 2.2% comparable to 3.6% reported from Ibadan [8]. This is a marked improvement from the previously reported mortality of 8–15.4% from different centers in developing countries [7,9,10,13,17].

The average cost of operative management of IS in this study is 260 USD; this is by far beyond the reach of many patients in low-income and middle-income countries like ours, where the majority live on less than 1 USD a day. It is worth mentioning that the majority of patients accessing healthcare services in developing countries pay out of their pocket as the Health Insurance schemes have limited coverage. In our study, only one patient was covered by the National Health Insurance Scheme during the study period. Early presentation and by extension nonoperative management of IS using ultrasound-guided hydrostatic saline enema reduction significantly reduced the total cost of care by almost half. There was an average of 21% ($50.0) additional charges incurred for each day's delay in presentation. The cost of operative procedures ($154 vs. $100, P=0.025) and drugs ($45 vs. $24, P=0.008) significantly contributed to total hospital charges. The length of stay was doubled for those who presented late (10.6 vs. 4.6 days, P=0.023). Given our limited resources, any amount of money saved from care of a patient will translate to judicious use of scarce funds.

This study is limited by all the flaws of retrospective studies and has limitation of not capturing all the cost of care of the patient because charges incurred that were not recorded cannot be accounted for. In addition, indirect costs of care like costs of facilities, maintenance, travels, missed school and missed work hours by a caregiver were excluded.

Conclusion

Delayed presentation of ISs translates to one-half total hospital cost and an extra week in hospital with utilization of limited resources and increased morbidity. Our study provides useful information on the estimated direct cost of treatment of IS putting the duration of symptoms in perspective. This we hope will encourage primary-care physicians, policy makers, and the public to develop strategies which focus on reduction of endemic late presentation in our settings.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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