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Özgün Araştırma / *Research Article*

Twenty Years of Experience in Management of Intussusception: Saline Enema Reduction with Ultrasound Guidance

İntususepsiyon Yönetiminde Yirmi Yıllık Deneyim: Ultrason Kılavuzluğunda Salin Lavmanlı Redüksiyon

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ABSTRACT

Objective: *Intussusception is a common condition involving infants and toddlers in which non-operative management plays an important role. The aim of this study was to present our 20 years of experience of saline enema reduction with ultrasound guide in management of intussusception.*
Methods: *A 20-year retrospective study was performed from January 1999 to December 2018 in Iran, Tabriz Pediatric Hospital. Candidates for inclusion in the study were children from 0 to 14 years old who were diagnosed with intussusception. Treatment option (surgical or saline enema reduction) was recorded along with primary and total success rate of saline enema reduction, early recurrence rate, surgery details and lead points.*
Results: *A total of 1376 patients with intussusception were studied. 112 patients experienced spontaneous reduction, and 1131 patients underwent ultrasonographic guided saline enema reduction. The primary attempt was successful in 76.8% of the patients (874 cases); however, in 8.4% of the patients (74 cases) recurrence was recorded during 48 hours after reduction. 264 patients were referred for second and or third reductions. The overall success rate of enema reduction was 79.9%. The mean age of the children with successful reduction was lower than the failed group (1.27±1.27 versus 1.63±1.05 years old; p=0.001). The success rate was significantly lower in patients with signs of currant jelly stool and palpable mass (p<0.001). 364 patients (26.5%) underwent surgery treatment. Leading points were recognized in 64 patients, and Meckel's diverticulum (63.24%) was the most common pathologic leading point.*
Conclusion: *Ultrasound-guided saline enema reduction is a feasible method and is suggested as an efficient and safe method for non-operative management of intussusception. Its success rate is high and could be increased further by proper case selection and repeated reduction attempts even in recurrent cases.*

Keywords: *Intussusception, Saline enema, ultrasonography*

ÖZ

Amaç: *İntususepsiyon, ameliyatsız tedavi yöntemlerinin önemli bir rol oynadığı bebekler ve küçük çocukları ilgilendiren yaygın bir durumdur. Bu çalışmanın amacı, intususepsiyonlu hastalarda ultrason kılavuzluğunda yapılan salin lavmanla redüksiyon konusundaki 20 yıllık tecrübemizi sunmaktır.*
Yöntem: *Ocak 1999 - Aralık 2018 tarihleri arasında 20 yıllık süreyi kapsayan bu retrospektif çalışma, Ocak 1999'dan Aralık 2018'e kadar Tebriz çocuk hastanesinde İran'da gerçekleştirildi. Çalışmaya dahil edilen adaylar, intususepsiyon teşhisi konan 0 ila 14 yaş arası çocuklardı. Tedavi seçeneği (cerrahi veya salin lavmanı redüksiyon), salin lavman redüksiyon birincil ve toplam başarı oranı, erken rekürrens oranı, ameliyat ayrıntıları ve sürükleyici noktaları ile birlikte kaydedildi.*
Bulgular: *İntususepsiyonu olan toplam 1376 hasta çalışıldı. 112 hasta spontan redüksiyon yaşadı ve 1131 hastaya ultrasonografi kılavuzluğunda salin lavman redüksiyonu uygulandı. Birincil girişim hastaların %76,8'inde (874 vaka) başarılı oldu; ancak, hastaların %8,4'ünde (74 vaka) redüksiyondan 48 saat sonra rekürrens kaydedildi. 264 hasta ikinci ve/veya üçüncü redüksiyon için önerildi. Lavman redüksiyon genel başarı oranı %79,9 idi. Başarılı redüksiyonu olan çocukların yaş ortalaması başarısız gruba göre daha düşüktü (1,27±1,27'ye karşı 1,63±1,05; p=0,001). Çilek jölesi dışkısı ve ele gelen kitle bulguları olan hastalarda başarı oranı anlamlı olarak daha düşüktü (p<0,001). 364 hastaya (%26,5) cerrahi tedavi uygulandı. Sürükleyici nokta 64 hastada fark edildi ve Meckel divertikülü (%63,24) en sık belirlenen patolojiydi.*
Sonuç: *Ultrason eşliğinde salin lavmanla redüksiyon, uygulanabilir bir yöntemdir ve intususepsiyonun ameliyat dışı yönetimi için etkili ve güvenli bir yöntem olarak önerilmektedir. Başarı oranı yüksektir ve tekrarlayan vakalarda bile uygun hasta seçimi ve tekrarlanan redüksiyon girişimleri ile daha da arttırılabilir.*

Anahtar kelimeler: *İntususepsiyon, Salin lavman, ultrasonografi*

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Introduction

Intussusception is the most common cause of bowel obstruction in infants and toddlers. The incidence of intussusception is approximately 1-4 per 1000 infants and children with male predominance [1,2]. In modern practice, non-operative treatment is preferred over surgical approaches for certain patients. However, there is a disagreement between centers using ultrasound guided saline enema, and fluoroscopic guided air enema [3,4]. Barium enema was once commonly used, but its application is now limited to few centers [1]. Long term experience in intussusception treatment using saline enema reduction would be helpful to determine the ideal approach for treatment.

In the pediatric surgical department of Iran, Tabriz University, a large number of patients are referred or admitted with a primary diagnosis of intussusception. Since 1999, the staff and equipment are available at all times for performing ultrasound-guided saline enema reduction as the non-operative treatment of intussusception. The aim of this study was to describe the 20 years of experience of our center in the treatment of intussusception and application of saline enema as the preferred reduction method for this condition.

Material and Methods

This study was a retrospective study approved by the Ethics Committee of Tabriz University of Medical Science (IR.TBZMED.REC.1399.042). Due to the retrospective nature of this study, there was no need for patient consent.

Patients: We reviewed the medical records of all patients with a final diagnosis of intestinal intussusception who were admitted to the referral center of Tabriz Pediatric Hospital over 20 years from January 1999 to December 2018. Data including demographic features (age, sex, body weight), disease presentations and symptoms, ultrasonographic reports, surgical procedural information, hospital stay, recurrence, and post-surgical complications. The diagnosis of intussusception was confirmed via an ultrasonographic study conducted by an experienced examiner according to the clinical guidelines for the diagnosis of acute intussusception. All patients with previously reported intussusception that were not confirmed by ultrasonographic examination were excluded. All patients with positive ultrasonogeraphic findings of intussusception were primarily hydrated with or without the administration of antibiotics and assessed for signs of peritonitis and sepsis. Prescribing antibiotics to the patients were done only in the cases with the systemic signs of general illness, fever or sepsis and also in the cases which required the second or third enema reduction following unsuccessful first try. Patients who had perforation signs and peritonitis and were unresponsive underwent urgent surgery. Others were planned for non-operative reduction by saline enema. Reduction was performed by a team consisting of a radiologist and a surgeon.

Enema procedure: A Foley catheter was used for administration of warm saline via anus of the patient, and the buttock was taped to prevent saline leakage. The continuous pressure of saline for enema was adjusted between 100 to 120 cmH_2O by saline height. Three separate attempts were done each one for up to 3 minutes. Sedative drugs were not used routinely for the patients undergoing enema reduction. The patients usually tolerated enema reduction with the presence and cooperation of their parents. In some rare cases syrup of chloral hydrate were given in the therapeutic dose. If new onset free fluid was detected during installation of saline, no more tries were done. If there wasn't any reduction in the size of intussusception in the term of huge intussusceptions the instillations are limited.

Since hydrostatic reduction with barium under fluoroscopic monitoring was traditionally accepted as the oldest and most widespread method, in the first year of the study, only a few cases were treated with this method. However, from the year 2000, saline enema reduction was the only preferred method for reduction in our center.

The success of saline enema reduction was determined by the disappearance of intussusception in the ultrasonographic study and the visualization of saline passing through the ileocecal valve or distention of ileum via saline.

Failed reduction was also defined as a remnant target or pseudo kidney sign as an indicator of intussusception mass, where saline could not pass from the cecum to the ileum. The second or third attempt for the reduction was done after reassessing the patients for the criteria of urgent surgery. In patients with probable underlying lead point according to clinical or radiological finding or atypical non ileocolic intussusceptions no more attempts for reduction were performed. These findings could be mentioned as concurrent prolonged or recurrent symptoms like intermittent rectal bleeding or abdominal pain and any other coexisting findings in sonographic study such as any mass or cystic lesion and unusual forms of intussusception including colocolic or huge extended intussusceptions

The second or third attempt for reduction was considered for other cases with the time interval ranging from 3 to 6 hours. Recurrence was defined as the occurrence of symptoms post-reduction with visualization of intussusception on follow-up ultrasonographic study. Unsuccessful reduction attempts were another indication for surgical treatment. The intussusception cases with non-diagnostic ultrasonographic study which were diagnosed during surgery were also included. All patients' medical records during the study were reviewed for early and late complications and further surgeries.

Statistical analysis: Statistical analysis was performed using IBM SPSS version 24.0. Group statistics were expressed as mean and standard deviation for categorical variables and percentage for descriptive analysis. Group comparisons were made using chi-square/Fisher exact test for categorical variables and a Student's t-test for continuous variables. $p \leq 0.05$ was considered significant in all statistical analysis. Logistic regression was performed to identify independent risk factors.

Results

A total of 1376 patients with the diagnosis of intussusception were admitted in our hospital during the 20-year study. The mean age of patients was 1.68±1.75 years old, ranged from 1 month to 14 years. The highest incidence rate was found to be in 6 months to 1-year-old age group. Age-depended distribution of patients is shown in Figure 1.

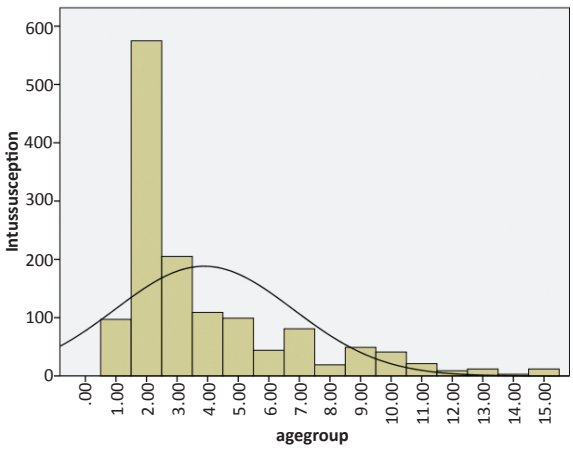

Figure 1. Age distribution of intussusception cases.

Male children accounted for 67.5% of the patients, and female children accounted for 32.5% of them. Male to female ratio was 2.07:1.

Only 7% (96 cases) of the patients had a previous history of recent viral infection in the last month.

In the age of 6 months to 3 years old, in which idiopathic intussusception is most common, 93.9% of cases had typical manifestations of colicky pain, and 6.1% did not present with typical pain.

Atypical manifestations were observed in 13.4% and 11.3% of patients with 0 to 6 months and more than 3 years of age, respectively. The mean age of patients presenting with abdominal pain was 2.17±2.00 years. Whereas, the mean age of patients with no presentation of abdominal pain was 0.90±0.77 ($p<0.001$).

Signs and symptoms of the patients are listed in Table 1.

Table 1. Frequency of signs and sumptoms in patients with intussusception.

Manifestation	Percentage (%)
Abdominal pain	61.6%
Vomiting	61.3%
Abdominal tenderness	29.0%
Diarrhea	21.4%
Currant jelly stool	24.6%
Dehydration	9.7%
Abdominal mass	8.2%
Tachycardia	3.5%

According to the feasibility, safety, and results, the ultrasonographic guided saline enema was selected as the treatment of choice in our center for last 20 years.

In 1327 patients, sonographic evaluations revealed the diagnosis of intussusception. Reported intussusception was a common ileocolic type in 94.9% of the patients. 1250 cases were candidate to non-operative treatment. 112 patients experienced spontaneous reduction of intussusception in the second ultrasonographic evaluation. 1131 patients underwent ultrasonographic guided saline enema, and 7 cases were treated by barium enema reduction.

Primary attempt to reduction was successful in 76.8% of patients (874 cases). In 74 patients (8.4%) recurrence of intussusception was recognized during the first 48 hours after reduction. From the unsuccessful reductions and early recurrent cases, 264 patients were suggested to undergo second or third reduction by saline enema. The success rate for the second or third reduction was only 36.7 % (99 patients). Overall success rate for non-operative treatment by enema reduction was 79.9% (909 from 1138 intussusceptions). We did not recognize any cases of obvious bowel perforations during the reduction procedure.

A total of 364 patients (26.5%) underwent surgical treatment. In 128 patients with primary diagnosis of intussusception, surgery was chosen as the initial treatment based on findings in physical examinations and imaging findings. Surgical treatment was also considered for 195 patients with frequent recurrences after the reduction and failure of non-operative treatments. Besides, in 49 patients with non-diagnostic sonographic findings (24 cases) or other primary diagnosis with atypical presentations (25 cases), intussusception was encountered during surgery.

The mean age of children with successful non-operative reduction was lower than children with failed reduction (1.27±1.27 versus 1.63±1.05 years old; p=0.001). The success rate in patients presenting with atypical pains was lower than patients with typical pains (p=0.001). Success rate was significantly lower in patient with currant jelly stool and palpable mass in physical examination. (p<0.001) (Table 2).

Surgical procedure was operative reduction without resection in 248 cases (67.4%). Resection and anastomosis were done as a result of bowel gangrene or detected lead point in 120 cases (32%).

The majority of patients with successful reduction (77.9%), stayed in hospital for less than 24 hours. Hospital stay time for 19.6% was 2 days. 2.5% of the patients were admitted in the hospital for 3 days. The mean hospital stay time for patients treated with surgery was 4.88±1.88 ranging from 1 to 18 days.

From 322 cases treated by surgical procedure, 59 cases (18.32%) had non-ileocolic intussusception, including ileoileal and jejunojejunal and colocolic intussusceptions. The type of intussusception determined by ultrasonography was incorrect in 8 cases (3.04%) of ileocolic intussusceptions. Moreover, from 38 (64.40%) of 59 cases with non-ileocolic intussusceptions were primarily diagnosed as common ileocolic type.

Pathologic findings considered as leading points were Meckel's diverticulum, polyps, colonic or ileal

Table 2. Comparison of characteristics and clinical factors in patients with successful and non-successful enema.

Variables	Successful enema reduction (N=909 (%))	non-successful enema reduction (N= 229(%))	X^2 value	P-value
Gender (male)	625 (68.75%)	146 (63.75%)	2.09	0.148
Positive history of intussusception	73 (8.03%)	18 (7.86%)	0.007	0.932
Typical pain	738 (81.19%)	125 (54.58%)	10.435	0.001
Vomiting	516 (56.76%)	178 (77.73%)	33.785	<0.001
Diarrhea	166 (18.26%)	63 (27.51%)	9.735	0.002
Currant jelly stool	171 (18.81%)	104 (45.41%)	70.641	<0.001
Abdominal tenderness	249 (27.39%)	70 (30.57%)	0.914	0.339
Abdominal mass	54 (5.94%)	38 (16.59%)	27.93	<0.001
Pathological lead point	6 (0.66%)	27 (11.79%)	80.479	<0.001

masses such as lymphoma, mesenteric, and duplication cysts (Table 3). Meckel's diverticulum was founded in 43 cases (63.24%) and considered as the most common pathologic leading point. One of the patients had Peutz-Jeghers syndrome with multiple gastrointestinal hamartomatous polyps. The mean age of patients with an underlying leading point was significantly higher than idiopathic cases being 3.50±3.41 and 1.59±1.55 years, respectively (p<0.001). Other concurrent findings include malrotation in 8 cases.

Table 3. Pathologic lead points associated with intussusception, divided in age groups.

	<3 yrs	≥3yrs	All cases
Meckel's diverticulum	30	13	43 (67.18%)
Benign polyps	7	6	13 (20.31%)
Peutz-Jeghers syndrome	0	1	1 (1.56%)
Lymphoma (Burkitt's, large cell, Non-Hodgkin lymphoma)	1	3	4 (6.25%)
Ectopic pancreatic tissue	1	0	1 (1.56%)
Mesentric cyst	0	1	1 (1.56%)
Duplication of cecum	0	1	1 (1.56%)
Total	39	25	64

In the follow-up period after surgery 29 cases with complications were encountered. Two cases with abdominal sepsis were expired, one during surgery and the other one 2days after following surgery. The summary of encountered complications is listed in the Table 4.

Table 4. Post-operative complications observed in follow up period.

	Frequency	Percent
Delayed bowel obstruction	15	4.12%
Anastomosis leak and abscess formation	2	0.54%
Prolonged ileus	1	0.27%
Dehiscence of abdominal wall	1	0.27%
Incisional hernia	4	1.10%
Wound infection	3	0.82%
Entero-cutaneous fistula needs colostomy	1	0.27%
Reoperation due to bowel gangrene	1	0.27%
Total	29	7.97%
Total surgeries	364	

Discussion

Intussusception is a common condition affecting infants and toddlers. A majority of cases do not have leading points and are classified as idiopathic intussusceptions. This condition is mostly attributed to hypertrophied Peyer patches within the bowel wall. Even though idiopathic intussusception can occur at any age but it is uncommon before 3 months and after 3 years of age. Secondary intussusception is caused by a pathologic lead point which facilitates trapping of proximal bowel into the distal bowel by peristaltic activity [1,5]. The incidence of intussusception as a result of lead points is suggested to increase with age. As described in our study, the most common pathologic lead point was Meckel's diverticulum following benign polyps and lymphoma involvement of intestinal wall. These findings were consistent with previous reports [6].

Atypical manifestations like atypical pain were frequent in patients in lower and upper limits of age. This may be due to obscure manifestation and widespread differential diagnosis in the first months of life. Also, secondary intussusception, which is more frequent in older patients, could interfere with typical manifestation in this group [7].

Many reduction methods have been introduced to avoid surgery for the treatment of intussusception. Barium reduction under fluoroscopic guide was the oldest and most widespread method for reduction. Pneumatic reduction under the fluoroscopic guide was introduced later with a worldwide application, especially following the study of Guo JZ et al. [2] (1986) on 6396 cases with a success rate of more than 90%.

Ultrasound-guided hydrostatic reduction by saline was first described in 1982 by Kim et al. [7] and suggested a non-invasive treatment of intussusception without any risk of radiation exposure. It has been demonstrated that ultrasound-guided enema reduction is as effective as fluoroscopic reduction without any risk of radiation for children. Reported complications such as bowel perforation are very uncommon following saline enema reduction and they usually do not alter the prognosis of surgical treatments [2,8].

The success rate for hydrostatic reduction of intussusception is reported to be variable from 80-97% in different studies [2,7,8]. In 2019, Xiaolong reported a success rate of 96.77% for hydrostatic reduction of intussusception in pediatric patients [9]. According to the findings of previous studies, success rates of this non-operative procedure are promising. However, it

seems that inclusion criteria and case selection have a great impact on the results of non-operative treatments, especially hydrostatic reduction. We suggest a low threshold for choosing hydrostatic reduction as the first treatment even in patients with borderline findings such as mild to moderate intra-abdominal fluid or prolonged intussusception without any definite signs of bowel perforation or peritonitis. Even though the success rate in this group of patients is relatively lower, due to minimal complications of hydrostatic reduction, its application could prevent unnecessary operations.

Although currant jelly stool, abdominal mass, and Dance sign are not indications for surgical management of intussusception, they are accompanied by lower success rates of reduction [9,10]. Moreover, in our study success rate for patients presented by vomiting was also relatively lower. Further comprehensive studies are suggested to study the risk factors of unsuccessful reduction in pediatrics.

Spontaneous reduction is possible in almost all types of intussusceptions [11]. It occurred in our study during the preparation time for a reduction in reported ileocolic or colocolic types or during the observation time for small bowel intussusceptions. It seems that ultrasonographic findings which are operative-dependent are less sensitive for proper differentiation between types of intussusception compared to post-surgical diagnosis. According to inaccuracies in ultrasonographic diagnosis of the origin of intussusception, we suggest that at least one attempt is made for a hydrostatic saline reduction in patients with suspicious small bowel intussusception in the age range of idiopathic intussusception, and without any recognized lead point.

Ultrasonographic diagnosis of successful reduction is doubtful in some cases, especially if residual edema is significant. In cases with suspicious ultrasonographic findings after reduction attempts, we decided to postpone surgical treatment and observe the patients using physical and ultrasonographic examinations for 4-6 hours after complete relief of clinical symptoms. Ultrasonographic findings resolved in some cases reducing unnecessary surgical explorations. This finding was described by Shehata as the psuedotarget sign [12].

2.19% of cases had malrotation during surgery as a concurrent finding. Whether malrotation can act as a lead point is unclear. The incidence of malrotation is estimated to be 1 in 6000 live births. However, an increased incidence of about 1-2% was reported in barium studies on autopsies seeming to be the actual incidence. Even though it is unclear if unusual intestinal fixation can act as a lead point for intussusception, the fact that colocolic intussuscetions which are less common in children are frequently seen in this condition could confirm this theory. Some limited reports suggest intestinal malrotation as a cause of intussusception and refer to the association as Waugh's syndrome [13,14]. In our study, from 8 patients with this association, 2 cases had colocolic, and one case had jejenojejenal intussusceptions that are less common in normal population. Further investigations are recommended to study the association in Waugh's syndrome.

As shown in the results, a positive history of previous enema reduction did not alter the success rate of further reductions [15,16]. We suggest surgical treatment only in cases with failure of saline reduction and suspicious lead points in several episodes of recurrence. In 2010, Niramis R et al. [17] studied appropriate management procedures for recurrent intussusception in 1340 patients and 108 episodes of recurrent intussusception. The success rates of barium enema and air enema were 92% and 96.2%, respectively, suggesting that the principle treatment of recurrent intussusception should be similar to that of primary intussusception [17].

The limitation of this study was the retrospective design and related potential selection bias. Multicenter, prospective studies are recommended for further evaluation of outcomes of ultrasound-guided saline enema and risk factors of unsuccessful reduction.

Ultrasound-guided saline enema reduction is an efficient and safe method for non-operative management of intussusception. It is a feasible method without radiation exposure and with a low complication rate. Its success rate is high and could be increased further by proper case selection, experienced radiologists, and repeated reduction attempts even in recurrent cases.

Ethics Committee Approval: Approval was obtained from the Ethics Committee of Tabriz University of Medical Science (IR.TBZMED.REC.1399.042)
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