ORIGINAL ARTICLE

Our treatment approaches in recurrent chronic intussusceptions

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ABSTRACT

BACKGROUND: Intussusception is the most common cause of intestinal obstruction between 6 months and 36 months of age. There is no defined etiology in at least 75–90% of patients. Recurrent intussusception occurs in 5–16% of all intussusceptions and the treatment strategy is controversial in this patient group. The treatment of continued recurrent intussusception is a challenging problem when no lead point is revealed despite recurrence.

METHODS: We aimed to review our 10 years of experience in recurrent intussusception and describe a new operative technique for recurrent intussusception cases without any lead points.

RESULTS: We, retrospectively, reviewed the data of patients with recurrent intussusception in our referral pediatric surgery clinic between 2007 and 2017. Ultrasound-guided hydrostatic reduction (UGHR) was performed on all patients. Surgery was performed on those patients who had findings of acute abdomen and complete intestinal obstruction or two failed attempts of UGHR for diagnostic purposes if a pathologic lead point was suspected based on patient findings and age. Laparoscopy or laparotomy was performed according to surgeon preference and experience. A total of 87 UGHRs were performed. Thirty-three patients were admitted to our clinic due to recurrent intussusception. The mean age was 12.75±14.14 (6–84) months, and 19 were male and 14 were female. Abdominal pain, agitation, and vomiting were common symptoms. UGHR was performed on all 33 patients on at least two different occasions. The time between the first and second UGHR treatments was 42.6±186.19 (0–899) days. The success rate of the second UGHR was 27 out of 33 patients (81.8%). Surgery was performed on six patients. Laparoscopy-assisted ileal folding and fixation to the cecal wall was performed on one patient with recurrent intussusceptions. Appendectomy was performed first, and then, ileal folding with cecal fixation was performed using 4/0 polyglactin sutures. The sutures were placed between the serosal layers of the adjacent terminal ileal loops and the cecal wall.

CONCLUSION: Surgeons should try to find permanent solutions for patients with multiple recurrent intussusceptions that are resistant to treatment. Surgical excision of the lead point will help prevent recurrent intussusception. Satisfactory results can also be obtained by UGHR even in patients with recurrences. Laparoscopy is helpful in diagnosis, detection of lead points, and treatment of irreducible intussusception. This new operative technique can be satisfactory for recurrent intussusceptions without any lead points.

Keywords: Idiopathic recurrent intussusception; ileal folding and cecal fixation; ileocolic pexy; ultrasound-guided hydrostatic reduction.

INTRODUCTION

Intussusception is the most common cause of intestinal obstruction between 6 months and 36 months of age. There is no defined etiology in at least 75–90% of patients and they are considered as idiopathic.[1,2] The pathology behind idiopathic intussusception is commonly thought to be caused by hypertrophy of Peyer's patches after viral infections.[3,4] Lead

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points have been found in 1.5–12% patients undergoing operative reduction and are common in patients aged below 3 months and over 5 years.[5–10] Ultrasound-guided hydrostatic reduction (UGHR) is a safe and effective method for treating intussusception.[11] Barium and air enemas are also useful in reducing uncomplicated intussusceptions.[12]

Recurrent intussusception occurs in 5–16% of all intussusceptions and the treatment strategy is controversial in this patient group.[9,10,13] There is no consensus for the optimal timing of surgery. In the literature, almost 25% of patients with intussusception need surgery eventually. Hsu et al.[14] proposed that surgical intervention should be considered in the third episode. Surgical procedures are performed for irreducible intussusception after failure of non-surgical reduction methods, in emergency cases or for diagnostic evaluation purposes. Detection of a lead point can be advantageous for treating the underlying cause in order to prevent recurrence. The main problem starts when no lead point is found after diagnostic investigations and operation. Recurrent intussusceptions without any lead points despite successful non-operative reductions are challenging for patients and medical teams. Novel treatment strategies are necessary for this condition.

The aim of this study is to evaluate the findings of patients with recurrent intussusception and describe our novel technique for treating patients who had recurrent intussusceptions without any lead points.

MATERIALS AND METHODS

Patients and Study Design

We, retrospectively, reviewed the data of patients with recurrent intussusception in our referral pediatric surgery clinic between 2007 and 2017. Intussusception was diagnosed by clinical presentation, physical examination, upright abdominal radiograph, and abdominal ultrasound. UGHR was performed on all patients.

Surgery was performed on those patients who had findings of acute abdomen and complete intestinal obstruction or two failed attempts of UGHR for diagnostic purposes if a pathologic lead point was suspected based on patient clinical findings. Laparoscopy or laparotomy was performed according to surgeon preference and experience. Demographic, clinical, and operative findings were recorded and evaluated retrospectively.

Inclusion and Exclusion Criteria

Patient records are searched for the diagnosis of intussusception. Pediatric patients aged 0–18 years who had undergone UGHR on at least two different hospital admissions were included in the study. All pediatric patients with only one episode of intussusception were excluded. Patients with acute abdomen who directly went to surgery were excluded from the study. Patients with ileoileal and colocolic intussusception were also excluded from the study.

The Novel Procedure

Before the operation, the suggested surgical procedure, other treatment options, and possible complications were explained by the medical team to the parents and informed consent was obtained. During laparoscopy, a 5-mm umbilical trocar and two additional 5-mm trocars, one in the left lateral quadrant and the other in the suprapubic area, were placed.

The exposure was initiated by examining the abdominal cavity, confirmation of diagnosis, and possible pathologic lead points (PLP). No PLP was found. The cecum and the distal ileum were delivered outside after the umbilical incision was enlarged. Appendectomy was performed first, and then, ileal folding with cecal fixation was performed using 4/0 polyglactin

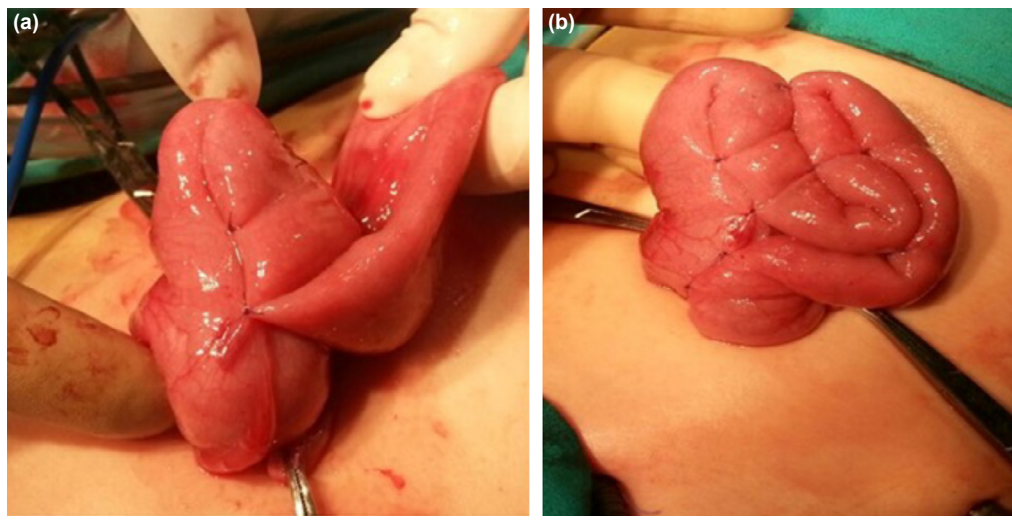

Figure 1. The view of the novel procedure, laparoscopy-assisted ileal folding and cecal fixation **(a)** ileal fixation on the cecum and **(b)** ileal folding.

sutures (Fig. 1). The sutures were placed between the serosal layers of the adjacent terminal ileal loops and the cecal wall. After fixation was complete, viability of the intestines was not affected. Intestinal passage was checked manually and no resistance was found.

"The novel procedure can be applied for patients with the recurrent idiopathic intussusception after no pathological findings could not find during the laparoscopic assessment."

Statistical Analysis

Mean, standard deviation, and ratios were used to analyze the data values. Kruskal–Wallis and Mann–Whitney U-tests were used to compare independent quantitative data. Spearman's correlation test was used for correlation analysis. The analysis was done in SPSS 22.0 software. The statistical significance rate was taken as $p<0.05$.

Ethics Approval and Consent to Participate

All procedures performed in study involving human participants were in accordance with the ethical standards of the Institutional and/or National Research Committee and with the 1964 Helsinki declaration and ethical approval was taken from both Local Ethics Committee (date 07.07.2020 and number 2877).

RESULTS

Thirty-three patients were admitted to our clinic due to recurrent intussusception. A total of 87 UGHR procedures were performed. Twenty-two patients had two, eight patients had three, one patient had four, one patient had seven, and one patient had eight recurrences. The mean age was 12.75±14.14 (6–84) months, and there were 19 male and 14 female. Abdominal pain, agitation, and vomiting were common symptoms. Visible blood in stool was seen in seven out of 33 patients and three had an abdominal mass on palpation. Increased white blood cell counts were observed in 73% of the patients, whereas 47% had elevated C-reactive protein values.

UGHR was performed on all 33 patients on at least two different occasions. The mean length of intussusception was 33.57±17.58 mm (15–103). The time between the first and second UGHR treatments was 42.6±186.19 (0–899) days. The success rate of the second UGHR was 27 out of 33 patients (81.8%). Surgery was performed on six patients. Manual reduction was performed on five patients and one was operated by the laparoscopic reduction of intussusception and Meckel's diverticulum resection.

In eight patients, UGHR was performed 3 times, and the mean length of intussusception was 34.8±18.7 (21–73) mm. The mean time period between the second and third UGHR procedures was 49.5±97.1 (1–299) days. The success rate of the third UGHR was six out of eight (75%) patients. A 7-year-old patient with failed reduction had Burkitt lymphoma on whom an ileal resection and anastomosis were performed. In another patient with a failed attempt on reduction through UGHR, the intussusception was manually reduced and no lead point was found on laparotomy. The intussusception in one patient with Henoch-Schönlein vasculitis was reduced 3 times (Table 1).

In one patient, UGHR was performed 4 times. The mean length was 36.7±8.2 (30–51) mm. The mean time between the episodes was 63.75±43.8 (35–137) days. The reductions were successful in all attempts. One patient underwent seven successful UGHR procedures.

Eight intussusceptions developed in one patient during a follow-up period of 34 months after he was 2 years old. He was admitted to our clinic from the first intussusception episode. Diagnostic laparoscopy was performed after the fifth intussusception; however, no lead points were found. Diagnostic laparoscopy was planned after the fourth episode, but it could not be performed due to an upper respiratory tract infection. A 22-mm intussusception was diagnosed at the eighth attack and could not be reduced by UGHR. On laparotomy, the hypertrophy of Peyer's lymph patches was considered as a lead point. The ileum was resected from 5-cm proximal to the ileocecal valve to 25 cm due to concerns regarding viability of the ileum. The patient needed a second operation due to obstruction with adhesions at post-operative day 11. There were adhesions between the appendix and the ileal anastomotic line. No new intussusceptions developed during the 3-years follow-up.

Seven intussusceptions developed in another patient in a period of 14 months. He was 9 months old at first admission.

Table 1. Data of patients with recurrent intussusceptions

Number of intussusceptions	Number of patients	Age (month)	Interval time between episodes (day)	Length of intussusception (mm)	UGHR success rate (%)
2	22	6–12	42.6±186.19 (0–899)	33.57±17.58 (15–103)	80.3
3	8	12–84	49.5±97.1 (1–299)	34.8±18.7 (21–73)	75
4	1	12	63.75±43.8 (35–137)	36.7±8.2 (30–51)	100
7	1	24	56.57±58.1 (3–180)	30.3±7.4 (22–43)	100
8	1	24	66.5±64.4 (3–210)	32.5±10.4 (22–57)	87.5

Table 2. The ages of patients with leading points

Age (month)	Leading point
84	Burkitt lymphoma
36	Cystic fibrosis
12	Meckel's diverticulum
24	Peyer's lymph nodes
72	Henoch-Schönlein Purpura

After the sixth UGHR performed due to intussusception, the novel operative procedure was carried out in the seventh episode. The laparoscopy revealed no lead points. Appendectomy was performed first. The distal ileum and the cecum were pulled out of the abdomen through an umbilical incision. A 20-cm segment of the terminal ileum was folded 4 times and fixed to the cecum through seromuscular absorbable sutures. These newly created intestinal loops were fixed to the cecum with one seromuscular suture, and then, the intestines and the cecum were placed back into their respective positions in the abdomen (Fig. 1).

Oral feeding was started on post-operative day 2 and the patient was discharged on day 4. The patient has been in follow-up for 4 years without any complications or recurrence. Another patient in whom the intussusception recurred 8 times has not had any complaints after the last UGHR.

No complications developed due to the UGHR interventions. All patients with recurrent intussusceptions were evaluated by a pediatric gastroenterologist for cystic fibrosis, inflammatory bowel disease, celiac disease, infectious diseases, and vasculitis. Cystic fibrosis was found in one patient. All patients were re-evaluated by a radiologist using ultrasound for lead points 2 weeks after hydrostatic reduction. Lead points were found in five out of 33 patients (15.1%) which consisted of a lymphoma, a cystic fibrosis, a Meckel's diverticulum, Peyer's lymph nodes, and Henoch-Schönlein Purpura (Table 2).

DISCUSSION

Nonsurgical reduction methods have been performed on the treatment of intussusception and widely used. Procedures that started with a barium enema continue successfully with hydrostatic reduction under ultrasound guidance and air enema today. UGHR has many advantages including avoidance of X-rays, cost efficiency, and accessibility as well as the fact that ultrasound can be helpful for detecting PLPs. However, the main disadvantage of UGHR is that pediatric surgeons would need radiologists or an ultrasound training. Ultrasound is commonly used by many clinicians now. In addition, UGHR is commonly accepted to render successful results for the treatment of cases with a first episode, and minimum complications are reported in the literature.[11] Depending on these results, UGHR is also accepted for use in recurrent intussusceptions. Some of the recurrences are due to insufficient reductions that may result from an inexperienced radiologist, fear of bowel perforation, or the nature of the disease depending on PLP or idiopathic intussusception. There is still no consensus for the number of episodes or timing of operation in recurrent intussusception.[14] However, the main challenge for clinicians continues to be recurrent cases despite lack of any PLPs in laparoscopy or laparotomy.[13] In addition, repeated reductions by enema and laparoscopy and the absence of PLPs have a discouraging effect on families and surgeons. In the literature, Kaiser et al.[15] compared air and saline enemas for the treatment of intussusception. In this study, air enema was found to be slightly more advantageous for cases with symptom presence longer than 24 h. However, Niramis et al.[13] reported no statistically significant difference in the intussusception reduction rates between barium and air enemas for non-operative treatment. In the latter study, the success rate of UGHR was higher than 80% in patients with two and more episodes, even if the patient had been operated before. Our success rate is slightly higher than what was reported in the literature. In the study of Kaiser et al.,[15] the success rate was 42–80%, and this lower success rate was attributed to delayed diagnosis. In addition, we have to note that after surgical manual reduction, the recurrence rate is lower than both enema methods. No recurrence was observed after bowel resection for intussusception in large series.[13,15–17] In our series, one patient was treated with UGHR despite a previous surgical manual reduction. UGHR can be useful in a patient with recurrent intussusception despite previous reductions by non-operative or operative methods.

Perforation of the bowel is a significant complication in non-operative treatment. Perforation rates of air or barium enemas by fluoroscopy or USG are 0.1–1.2% and 0.5–2.5%, respectively.[10,11,18,19] In our series, no bowel perforation or major complications occurred. We believe that during intervention, bowel diameter should be monitored by USG intermittently.

The overall recurrence rate is 8.5–12.7% in all patients with intussusception. Meckel's diverticulum has been shown as a common cause for recurrence.[14] PLP such as polyps, lymphadenopathy, lymphoma, appendix, duplication, rotavirus vaccination, and vasculitis have been described in the literature as other causes of recurrence[8,20–28] PLP is generally diagnosed during operation due to unreduced intussusception at a rate around 8–12%.

In the literature, there is no clear consensus about the timing of surgery after intussusception episodes. Different studies suggest attempting non-operative methods between the second and fifth episodes unless a PLP is detected or irreducible intussusception is encountered.[14] This uncertain process can be worrisome for patients' families and medical teams. Moreover, the main question is how to manage these patients when there is no clear lead point present. Usually, lead points cannot be determined in patients with intussusception and in some patients, recurrent attacks can be seen without any

detected PLP. Multiple interventions by enema may involve an increased possibility of cumulative complications and be disappointing for the family and the surgeon.

Glucocorticoids were used for treatment in a patient with multiple recurrences. It was proposed that glucocorticoids diminished recurrence due to a reduction in lymphoid hyperplasia.[26,28]

The literature involves some reports regarding the fixation of the ileal segment and the cecum to peritoneal tissues for such rare condition. Burrington first described the ileocolic pexy technique for the prevention of recurrent intussusception in childhood.[29] The laparoscopic approach has generally been accepted for reducing intussusception and searching for a lead point. In a large case series, laparoscopic ileocolic pexy for ileocolic intussusception with multiple recurrences was explained for this patient group. This procedure was used after more than two recurrences without peritonitis or septicemia. Under this technique, the terminal ileum was sutured to the lateral peritoneum after appendectomy.[29] In our patient, the lateral peritoneum was checked for fixation, but no suitable regions were detected as the estimated suturing area was close to the iliac vessels.

Simple manual reduction of ileocolic intussusception with ileopexy or segmental ileal resection is described surgical procedure for recurrent intussusception.[29,30] In one patient, partial ileal segment resection including the Peyer's node and appendectomy was performed. Recurrence was not seen in this patient. The patient was operated due to the adhesion of the stump of the appendix to the anastomotic line. however, this complication can develop after any abdominal operation. The ileum has important duties in the digestive system, especially with regard to Vitamin B12 absorption. In general, bowel resection should be avoided as much as possible. Our novel procedure may be suitable for patients with multiple recurrences and no lead points, especially when fixation to the peritoneum is not deemed possible by the surgeon. Multiple UGHR procedures, hospital admissions, and family and surgeon frustration may lead to a decision for a new definitive solution. Limited solutions are available in serious cases such as ileocolic pexy to the peritoneum. However, there is no exact procedure for idiopathic recurrences in the literature. We performed ileal folding and fixation to the cecum for such conditions. We were very selective about performing this procedure, because we know that recurrent intussusception may be a temporary condition and depend on patient's age. The described approach is easy to apply by laparoscopic assistance, less invasive compared to open procedure, and does not require suturing close to the iliac vessels.[31] In addition, both procedures can explain how prevention of recurrence occurs. First, we performed controlled adhesions with sutures in the terminal ileum. The adhesions prevent easy movement of the distal ileum to the cecum. A fixated cecum with ileal folding can move together like a pendulum clock according to peristaltism. As a result, the novel approach can be effective for a long time without any serious complications. If we had done the literature review before, we could have also considered this new method for other patients. Since we have now applied this method and observed its success, if any multiple recurrent intussusception patients under our follow-up refer to us again for recurrence, we may consider this new method of operation upon detailed family consent. One patient with the ideal folding and fixation to cecum procedure is not enough to solve the idiopathic recurrent intussusception, but it can be good alternative for these patients.

Conclusion

It is known that invaginations are rarely seen except in infancy and early childhood. Surgeons should try to find permanent solutions for patients with treatment-resistant multiple recurrent intussusceptions. The presence of a leading point may indicate curative treatment. Even in recurrent patients, satisfactory results can be obtained with UGHR. Laparoscopy is helpful in the diagnosis of irreducible intussusception, detection, and treatment of precursors. The treatment of idiopathic multiple intussusceptions can be challenging for clinicians despite all the interventions. There are no clear explanations about the timing of diagnostic laparoscopy and which surgical procedure can be performed. Although surgery performed in a single patient does not reflect evidence, we think that it can be considered as a treatment option in cases of recurrent intussusception without a lead point. Therefore, this new procedure may be an alternative surgical option for challenging cases of idiopathic multiple intussusceptions without any proven leading edge. Larger series are needed to see the effectiveness of the method and to determine the indication criteria.

Ethics Committee Approval: This study was approved by the University of Health Sciences, Şişli Hamidiye Etfal Training and Research Hospital SUAM, Clinical Research Ethics Committee (Date: 07.07.2020, Decision No: 2877).

Peer-review: Internally peer-reviewed.

Authorship Contributions: Concept: M.A.; Design: M.D.; Supervision: M.A.; Resource: M.K.; Materials: N.S.; Data: M.D.; Analysis: A.İ.D.; Literature search: M.D.; Writing: A.Ü.; Critical revision: N.S.

Conflict of Interest: None declared.

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ORİJİNAL ÇALIŞMA - *ÖZ*

Tekrarlayan kronik invajinasyonlarda tedavi yaklaşımlarımız

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AMAÇ: İnvajinasyon, 6 ay ile 36 ay arasında en sık görülen intestinal obstrüksiyon nedenidir. Hastaların %75–90'ında tanımlanmış bir etiyoloji yoktur. Tüm invajinasyonların %5–16'sında tekrarlayan invajinasyon görülür ve bu hasta grubunda tedavi stratejisi tartışmalıdır. Tekrarlayan invajinasyonun tedavisi, herhangi bir lead point bulunamadığında zorlu bir problemdir.

GEREÇ VE YÖNTEM: Tekrarlayan invajinasyondaki 10 yıllık deneyimimizi gözden geçirmeyi ve herhangi lead point olmayan tekrarlayan invajinasyon olguları için yeni bir ameliyat tekniği tanımlamayı amaçladık.

BULGULAR: 2007–2017 yılları arasında çocuk cerrahisi kliniğimizde tekrarlayan invajinasyonlu hastaların verilerini geriye dönük olarak inceledik. Tüm hastalara ultrason eşliğinde hidrostatik redüksiyon (UGHR) uygulandı. Akut karın ve tam bağırsak tıkanıklığı bulguları olan veya tanı amaçlı iki başarısız UGHR girişimi olan hastalara, bulgulara ve yaşına göre patolojik bir lead pointten şüpheleniliyorsa cerrahi uygulandı. Cerrahın tercihi ve tecrübesine göre laparoskopi veya laparotomi uygulandı. Toplam 87 UGHR gerçekleştirildi. Otuz üç hasta tekrarlayan invajinasyon nedeniyle kliniğimize başvurdu. Yaş ortalaması 12.75±14.14 (6–84) ay olup, 19'u erkek, 14'ü kadındı. Karın ağrısı, ajitasyon ve kusma yaygın semptomlardı. Otuz üç hastanın tamamına en az iki farklı durumda UGHR uygulandı. Birinci ve ikinci UGHR tedavileri arasındaki süre 42.6±186.19 (0–899) gündü. İkinci UGHR'nin başarı oranı 33 hastadan 27'si (%81.8) idi. Tekrarlayan invajinasyonları olan bir hastaya laparoskopi yardımlı ileal katlama ve çekum duvarına fiksasyon uygulandı. Önce apendektomi, ardından 4/0 poliglaktin sütürlerle çekal fiksasyon ile ileal katlama yapıldı. Dikişler, komşu terminal ileal ansların serozal tabakaları ile çekal duvar arasına yerleştirildi.

TARTIŞMA: Tedaviye dirençli çoklu tekrarlayan invajinasyonları olan hastalarda kalıcı çözümler bulmaya çalışılmalıdır. Lead pointin cerrahi olarak çıkarılması, tekrarlayan invajinasyonu önlemeye yardımcı olacaktır. Yineleyen hastalarda bile ultrason eşliğinde hidrostatik redüksiyon ile tatmin edici sonuçlar elde edilebilir. Laparoskopi, tekrarlayan invajinasyonun tanısında, lead point saptanmasında ve tedavisinde yardımcıdır. Bu yeni ameliyat tekniği, herhangi bir lead point olmayan tekrarlayan invajinasyonlar için tatmin edici olabilir.

Anahtar sözcükler: İdiyopatik tekrarlayan invajinasyon; ileal katlanma ve çekal fiksasyon; ileokolik peksi; ultrason eşliğinde hidrostatik redüksiyon.

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