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Successful treatment of intussusception by hydrostatic reduction in pediatric patients: Is everything okay?

Pedriatrik hastalarda hidrostatik redüksiyon ile başarılı intususepsiyon tedavisi: Her şey tamam mı?

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Abstract

Aim: Intussusception, which is defined as telescopic insertion of proximal bowel segment into distal bowel segment, can be cured completely with surgical intervention. Intussusception can be successfully treated by ultrasound guided hydrostatic reduction (USGHR) if there is no necrosis or perforations of intestines. However, misdiagnosing or omitting secondary conditions which can be seen together with intussusception leads to an inevitable rise in morbidity. In this study, we would like to present a retrospective review of the intussusception patients which developed complications due to misdiagnosis within a pediatric surgery clinic in terms of diagnosis and treatment.

Methods: 12 patients who were treated for intussusception using USGHR between May 2014 and September 2017 in Van Yüzüncü Yıl University Faculty of Medicine Pediatric Surgery Department were retrospectively reviewed for missed conditions and coincidental pathologies. The data about these case series such as age, sex, patient symptoms, diagnosis and treatment methods, complications and hospitalization periods were evaluated.

Results: 12 (5 Female – 7 Male) patients, who were diagnosed with invagination with complaints of abdominal pain, refractory emesis, crying attacks, bloody stool and abdominal distension that treated with USGHR with a mean age of 34 (Range 6 – 98) months, showed a worse clinical prognosis due to missed secondary conditions. Missed secondary pathologies included appendicitis (3 cases), lymphoma (1 case), Meckel diverticulitis (1 case), appendiceal intussusception (3 cases), acute gastroenteritis (3 cases) and Henoch-Schonlein Purpura (1 case). The patient with Henoch-Schonlein purpura diagnosis was treated with USGHR in combination with corticosteroids and all the other cases required open surgery. Follow-up of the patient with Henoch-Schonlein purpura is ongoing whereas all the other cases were treated successfully.

Conclusion: Although most intussusception cases are successfully treated with non-surgical USGHR treatments in our pediatric surgery clinic, missing the conditions that are seen with invagination causes an increase in surgical intervention rates, morbidity rates and medical costs. The main challenge for pediatric surgeons in invagination cases caused by pathological leading point conditions is the possibility of missing the actual underlying disease which caused the invagination following a successful USGHR after target-sign is detected. Although ultrasound and computed tomography studies might be helpful in preliminary diagnosis, it must be kept in mind that an actual diagnosis can only be done with surgery in some cases.

Keywords: Misdiagnosis, Intussusception, Ultrasound guided hydrostatic reduction

Öz

Amaç: İntususepsiyon, proksimal bağırsak segmentinin distal bağırsak segmentinin içine teleskopik olarak girmesi olarak tanımlanır, kesin tedavisi cerrahidir. Bağırsaklarda nekroz veya perforasyon yoksa Ultrason eşliğinde Hidrostatik redüksiyon (USGHR) ile başarılı bir şekilde tedavi edilebilir. Bununla birlikte, intususepsiyon ile görülebilen ikincil anomalilerin yanlış tanı alması veya atlanması morbiditede kaçınılmaz bir artışa yol açar. Bu çalışmada, bir çocuk cerrahisi kliniğinde tanı ve tedavi açısından yanlış tanıya bağlı komplikasyonlar gelişen intususepsiyon hastalarının retrospektif olarak gözden geçirilmesini sunmak istedik.

Yöntemler: Mayıs 2014 ve Eylül 2017 arasında Yüzüncü Yıl Üniversitesi Çocuk Cerrahi Kliniği'nde intususepsiyon tanısı alan ve USGHR ile tedavi edilen 12 hasta ıskalanmış tanılar ve ek patolojiler açısından retrospektif olarak değerlendirildi. Bu olgu serisindeki hastaların yaşı, cinsiyeti, semptomları, uygulanan tanı ve tedavi yöntemleri, komplikasyonlar ve hastanede kalış süreleri değerlendirildi.

Bulgular: Ağrı, kusma, ağlama atakları, dışkıda kan ve abdominal distansiyon şikayetleri ile başvuran ve USGHR ile tedavi edilen ve ortalama yaşları 34 (Aralık 6 - 98) ay olan 12 (5 kadın - 7 erkek) hasta, atlanmış sekonder durumlara nedeniyle daha kötü bir klinik prognoz gösterdi. apandisit (3 olgu), lenfoma (1 olgu), Meckel divertikülit (1 olgu), apendikal intususepsiyon (3 olgu), akut gastroenterit (3 olgu) ve Henoch-Schonlein Purpura (1 olgu) atlanmış sekonder patolojilerdi. Henoch-Schonlein purpura tanısı olan hasta USGHR ve kortikosteroidler ile birlikte tedavi edildi ancak diğer tüm olgular açık cerrahi gerektirdi. Henoch-Schonlein purpurası olan hastanın takibi devam ederken, diğer tüm olgular başarıyla tedavi edildi.

Sonuç: Çocuk cerrahisi kliniğimizdeki invajinasyon hastalarının çoğu USGHR ile cerrahi yapılmadan başarılı bir şekilde tedavi edilmesine rağmen, sekonder patolojiler gözden kaçırıldığında cerrahi girişim endikasyonu, artan morbidite ve medikal maliyetler kaçınılmazdır. Lead point'in mevcut olduğu invajinasyon vakalarında, bu hastalar için en önemli dezavantaj, ultrasonografide target sign görüntüsünün saptanmasını takiben yapılan başarılı bir USGHR ye rağmen alta yatan patolojinin ıskalanmasıdır. Ultrasonografi ve bilgisayarlı tomografi çalışmaları ön tanı için yararlı olsa da, gerçek bir teşhisin sadece bazı durumlarda cerrahi ile yapılabileceği akılda tutulmalıdır.

Anahtar kelimeler: Yanlış teşhis, İntususepsiyon, Ultrason eşliğinde hidrostatik redüksiyon

Introduction

Intussusception, which is the most common cause of intestinal obstructions seen in children between 4-10 months of age, can either be idiopathic or caused by a variety of conditions such as Meckel diverticulum, intestinal duplication, benign polyps, malignant lymphoma, Peuts-Jeghers syndrome, mesenteric cysts, intestinal wall hematoma of hemophilia, allergic purpura or hamartoma [1-3]. In idiopathic intussusception, the first line of non-surgical treatments are the pneumatic or hydrostatic reduction when there is no intestinal necrosis or perforations are present [1]. As the incidence rate of intussusception with a secondary condition is around 6%, surgical treatment is indicated in most of those cases [2,4]. In those cases, severe or even fatal complications such as bowel perforations or peritonitis can be seen due to the difficulties in the treatment of the conditions [2,5]. In intussusception diagnosis, ultrasonography (USG) has an almost 100% sensitivity and specificity. However, in complicated intussusception cases with various pathological anomalies ultrasonography or computed tomography (CT) does not show the similar success rates [2,3,6]. In those cases, secondary anomalies which can be seen with intussusception due to wrong or incomplete diagnosis can easily be missed.

In patients with intussusception, when the reduction treatment whether using gas or liquid enemas is unsuccessful, the patients can be taken into emergency surgery [2]. This study's main objective is to assess the patients who developed complications due to missed additional conditions with intussusception in terms of diagnosis and treatment.

Materials and methods

In our clinic, ultrasound guided hydrostatic reduction (USGHR) is routinely used for treatment of intussusception with exception of these conditions such as leading point, bowel necrosis and peritonitis. All patients received intravenous 0.9% Normal Saline 20 mg/kg/hour and midazolam was given as a premedication. Due to perforation risks, antibiotic prophylaxis was given in a single dosage of 50 mg/kg cephazolin and 30mg/kg metronidazole. With the patient laying in right lateral position a 14-18 Fr Foley's catheter was introduced into the anal canal and the balloon was inflated with 15-25 ml of saline. Foley's catheter size was chosen according to patient body size. Then the balloon of the catheter was placed in anal seal to avoid leaking back of the fluid. The on call radiologist used high resolution ultrasound IU22 (Philips, Netherlands) to image the intussusception. A clinician from Pediatric Surgery department remained in attendance in the suite.

Normally USGHR is ineffective in existence of leading point. Uncommonly in the proper use of USGHR can solve invaginated segment while there is leading point or additional pathologies. In this retrospective study; twelve patients that treated with USGHR with misdiagnose conditions and coincidental pathologies in Van Yuzuncu Yil University Faculty of Medicine Pediatric Surgery Department from May 2014 and September 2017.

Patients' age, gender, symptoms, treatment techniques, complications and hospitalization periods were retrospectively noted.

Results

The study included 12 patients (5 female, 7 male) with a mean age of 34 (Range: 6-84) months. The patients all presented to the clinic in 72 hours right after the first symptoms are seen. Patients had at least one of the following symptoms which are abdominal pain (12 cases), emesis (9 cases), crying jags (8 cases), blood in the stool (4 cases) and abdominal distension (2 cases). In total abdominal x-rays, it was seen that the colonic gases built up in the upper left quadrant of the abdomen in 6 patients. Following the detection of target-sign in USG, all patients had a pre-diagnosis of intussusception. The patients were treated by using USGHR. After the confirmation of the successful treatment with USG, the patients were discharged.

During post-op follow-up period, a number of patients presented to the clinic again after 2 to 32 days following discharge with complaints of loss of appetite, emesis, acute abdominal pain, abdominal distension and bloody stools. 3 patients with repeated complaints of loss of appetite, emesis and abdominal distension following 2-5 days after discharge were taken into emergency open surgery and perforated appendicitis was diagnosed. In 2 patients who presented to the clinic on 16th and 32nd days of successful USGHR treatment with acute abdomen were diagnosed with Meckel diverticulitis (1 case) and Burkitt lymphoma (1 case) (Figure 1, 2) during surgery.



Figure 1: Invaginated ileo-ileal segment

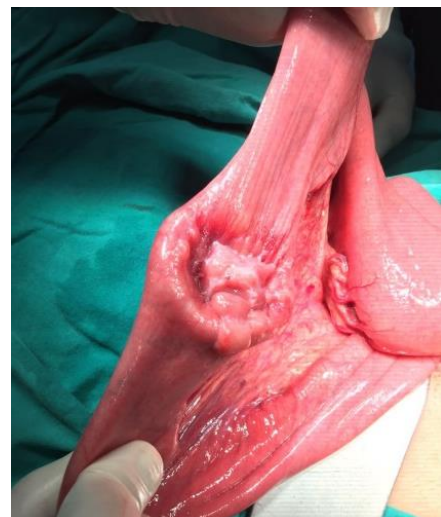


Figure 2: Burkitt Lymphoma as a leading point

In 3 patients with continued complaints of intermittent abdominal pain and emesis were successfully treated with incidental appendectomy although repeated intussusception or leading points were not detected. In three patients with suspected leading point following a repeated invagination were taken into surgery and the patients were diagnosed with acute gastroenteritis instead of leading point. A patient who presented to the clinic with rectal bleeding, palpable purpura in lower extremity and repeated intussusception was treated with USGHR in combination with corticosteroids and target-sign appearance disappeared after the treatment in follow-up USG. This patient was taken into follow-up for Henoch-Schonlein purpura. And all other patients made a successful recovery. The patients were briefly documented on the table 1.

Table 1: Demographic data of patients with misdiagnose conditions, treatment method and second application time to hospital after discharge

Age(months) / Gender	Misdiagnose Conditions	Treatment	Second admission to the hospital after discharge
24/F	Acute Appendicitis	Laparotomy	2 Days
36/M	Acute Appendicitis	Laparotomy	2 Days
24/M	Acute Appendicitis	Laparotomy	3 Days
38/M	Meckel Diverticulum	Laparotomy	16 Days
70/M	Henoch-Schonlein Purpura	Steroid	3 Days
10/F	Acute Gastroenteritis	USGHR	2 Days
14/M	Acute Gastroenteritis	USGHR	1 Day
13/F	Acute Gastroenteritis	USGHR	2 Days
40/M	Appendiceal Intussusception ?	Appendectomy	3 Days
58/F	Appendiceal Intussusception ?	Appendectomy	1 Day
19/F	Appendiceal Intussusception ?	Appendectomy	3 Days
62/M	Burkitt's Lymphoma	Excision of the mass	32 Days

F: Female, Male: Male

Discussion

Additional conditions such as appendicitis, lymphoma, Meckel diverticulitis, appendiceal intussusception, acute gastroenteritis or Henoch-Schonlein purpura can be missed during non-surgical treatment of invagination patients with or without leading points. Those missed cases due to wrong or incomplete diagnosis causes an increase in morbidity, medical costs and hospitalization periods of the patients. This study gives out a guideline on the diagnosis and treatment for the pediatric surgeons who come across with such cases.

Intussusception is second most common reason for emergency intervention following acute appendicitis in pediatric surgery clinics and is the most common cause for intestinal obstructions seen in children between 4-10 months [1,7]. Intussusception is seen in 1-4 in every 1000 live births in developed countries whereas the incidence rate is slightly higher in developing countries [1,7]. The most common symptoms include refractory emesis, intermittent abdominal cramps and pain with varying degrees of severity and bloody stools [1,8]. Intussusception usually has an idiopathic nature but in about 6% of the cases, an underlying pathological leading point is present [1,2]. In idiopathic intussusception diagnosis, ultrasonography has an almost 100% sensitivity and specificity rate [6]. Non-surgical interventions such as hydrostatic, pneumatic or external manual reduction techniques are the first line of choice in treatment in cases without bowel necrosis or perforation [6]. In

the event of complication development following an unsuccessful non-surgical procedure, surgical interventions are indicated as the most reliable method [6].

The main challenge for pediatric surgeons in intussusception is the diagnosis and treatment of invaginations caused by pathological leading points [1,2,5,10]. In those patients, USG has a 75% sensitivity and specificity rate whereas CT has 50% [2]. This situation is the main reason of missing the underlying anomaly which causes invagination in the first place or additional anomalies which can be seen with invagination. Chang's [11] study reported a non-surgically treated case diagnosed with intussusception. The patient's condition got worse and patient was taken into surgery. While the surgeons were expecting to find intussusception-related complications, they detected a missed diagnosis of perforated appendicitis. Another study done by Newman [12] on 6 cases reported that ultrasound images of a perforated appendicitis can mimic intussusception. An interesting point we found out was the absence of gas build-up in upper left quadrant and crying attacks in invagination cases with appendicitis. The most important disadvantage in those conditions for pediatric surgeons is the possibility of missing an underlying acute appendicitis diagnosis following a successful reduction of invagination, which the appendicitis is the leading point. Good USG results following USGHR for both clinical and control purposes can be deceiving for the surgeon. Another possible misdiagnosis is appendiceal intussusception which has the common clinical and radiological findings of both appendicitis and intussusception [13,14]. Target sign, which is typical in USG of intussusception, can be detected in this condition. They are usually spontaneously resolved or USGHR is used to reduce it easily. Due to the possibility of frequent recurrences, we suggest appendectomy in cases which require surgery for intussusception.

Intussusception rates peak during summer and winter months when respiratory and gastrointestinal infections are frequent [15]. Acute gastroenteritis following a successful reduction can cause recurrent intussusception. For this reason, we suggest a repeated USGHR in recurrent intussusception when there is no clear surgical indication and suspicion about leading points. In the literature, there are intussusception cases that are reported to have leading points of Henoch-Schonlein purpura and Burkitt lymphoma [16-18]. Especially in our patient with Burkitt lymphoma, we find it really interesting that the patient showed both good clinical and ultrasonography results following a successful USGHR. In intussusception cases with Henoch-Schonlein purpura, adding steroids to USGHR treatment can prevent recurrences.

Limitations of our study are its retrospective design, the low number of patients and the short follow-up period after USGHR treatment of intussusception. A prospective study involving a large number of patients may provide early detection of misdiagnosis and coincidental pathologies.

Conclusion

Although most intussusception cases are successfully treated with non-surgical USGHR treatments in our pediatric surgery clinic, missing the conditions that are seen with intussusception causes an increase in surgical intervention rates, morbidity rates and medical costs. The main challenge for

pediatric surgeons in intussusception caused by pathological leading point conditions is the possibility of missing the actual underlying disease which caused the intussusception following a successful USGHR after target-sign is detected. Although USG and CT studies might be helpful in preliminary diagnosis, it must be kept in mind that an actual diagnosis can only be done with surgery in some cases.

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