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**The importance of prompt diagnosis and early surgery in
Waugh syndrome**

Значај правовремене дијагнозе и ране хируршке интервенције код
Waugh синдрома

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The importance of prompt diagnosis and early surgery in Waugh syndrome

Значај правовремене дијагнозе и ране хируршке интервенције код *Waugh* синдрома

SUMMARY

Introduction Waugh syndrome (WS) is a rare condition characterized by association of intussusception and gut malrotation. Due to non-operative management for intussusception in certain number of patients, it still remains undiagnosed. So far, only less than hundred cases have been reported in literature, most of them treated surgically, with good outcome and no complication if surgery is done timely after symptoms appeared.

Case presentation We present a five month old infant which was admitted due to vomiting, blood and mucus in stool. Clinical findings revealed abdomen which was painful on palpation, without distension and abdominal mass. Blood and mucus were noted on rectal examination. After native X ray, abdominal ultrasound was done and confirmed intussusception. After unsuccessful attempt of hydrostatic reduction, ultrasound was repeated, the target sign persisted, this time accompanied by obvious “whirlpool sign” and inverse ratio of superior mesentery artery and vein. Infant underwent emergency surgery, and bowel resection with ileo-colic T-T anastomosis. Recovery was complete after two weeks.

Conclusion Waugh syndrome should be considered whenever hydrostatic reduction fails in a patient with previously diagnosed intussusception, and ultrasound verification of the superior mesenteric artery and vein relationship should be insisted upon to confirm the diagnosis off associated malrotation. In proven Waugh syndrome, timely surgical intervention is indicated to prevent bowel resection.

Keywords: Waugh syndrome; intussusception; malrotation; ultrasound

САЖЕТАК

Увод *Waugh* синдром (BC) је ретко стање које карактерише повезаност инвагинације и малротације прева. Због неоперативног лечења инвагинације, код одређеног броја пацијената и даље остаје недијагностикован. До сада је у литератури пријављено само нешто мање од стотину случајева, већина њих је лечена хируршки, са добрим исходом и без компликација ако се операција изврши благовремено након појаве симптома.

Приказ болесника Представљамо одојче узраста пет месеци које је примљено због повраћања, крви и слузи у столицу. У клиничком налазу абдомен је био болан на палпацију, без дистензије и палпабилних тумефакција. Приликом ректалног прегледа, примећени су трагови крви и слузи. Након нативног рендгенског снимка трбуха, урађен је ултразвук абдомена којим је потврђена инвагинација. Након неуспешног покушаја хидростатске редукције поновљен је ултразвук, „знак мете“ је перзистирао, овог пута праћен очигледним „знаком вртлога“ и инверзним односом горње мезентеричне артерије и вене. Одојче је подвргнуто хитној операцији и ресекцији прева са илео-количном Т-Т анастомозом. После две недеље дете се потпуно опоравило.

Закључак О *Waugh* синдрому треба размишљати сваки пут када не успе покушај хидростатске дезинвагинације код пацијента са претходно дијагностикованом инвагинацијом и треба инсистирати на ултразвучној верификацији односа горње мезентеричне артерије и вене како би се потврдила дијагноза удружене малротације. Код доказаног *Waugh* синдрома индикована је правовремена хируршка интервенција како би се спречила ресекција прева.

Кључне речи: *Waugh* синдром; инвагинација; малротација; ултразвук

INTRODUCTION

Waugh syndrome (WS) is a rare condition characterized by association of intussusception and gut malrotation [1]. Brereton and colleagues called this association “Waugh syndrome” after George E Waugh, who was the first to describe it in 1911 [2]. Only one prospective study was known so far to investigate high incidence of malrotation among patients with intussusception, and 18 out of 49 patients with intussusception had malrotation [3]. Incidence of WS is less than

1 % in the pediatric population [4, 5]. So far, fewer than 100 cases have been described worldwide [6]. It mostly occurs in the pediatric population, in the age range of 3 months to 3 years, some cases have recently been reported in adults [7].

We present a case of an infant with this association which was timely diagnosed and treated adequately.

CASE REPORT

A five-month-old girl was referred to the pediatric emergency department for suspected intussusception. Her medical and family history was normal, as were her obstetric, perinatal, and postnatal history. General complaints were vomiting, blood and mucus in a stool. Symptoms began the day before hospitalization. The first symptom was non-projectile bilious vomiting after a meal. On the same day, a few hours later, the next symptom appeared: blood and mucus in the stool which became increasingly pronounced the next day. The last passage of a stool was on the day of admission, and it contained a large amount of gelatinous bloody mucus.

On admission, physical examination revealed a moderately dehydrated, alert baby with rapid reactions. The abdomen was painful on palpation, without distension and abdominal mass. Blood and mucus were noted on rectal examination. The complete blood count was orderly. Inflammation parameters were slightly elevated (CRP was 27.4mg/L). Upright abdominal X ray showed irregular bowel aeration with air-fluid levels in the mid-portion of abdominal cavity (Figure1). Ultrasound confirmed intussusception with reactive, mesenteric lymph nodes and free fluid. Target sign was located approximately in the para-umbilical region, slightly on the right (Figure 2). An attempt of hydrostatic reduction was carried out in the next half an hour,

but unsuccessfully. Then ultrasound was repeated, the target sign persisted, this time accompanied by obvious “whirlpool sign” and inverse ratio of superior mesentery artery and vein (Figure 3). After that, a proximal gastrointestinal study was conducted, and revealed abnormal position of the duodenum, and stop in passage of contrast caudally.

Diagnosis of WS was confirmed, and emergency surgery was indicated. Intravenous resuscitation was administered, antibiotics parenteral (III generation of cephalosporins) as well as pain medications, and gastro-protective therapy. An nasogastric (NG) tube (8Ch) was placed.

The infant was admitted to the hospital 30 hours after first symptoms and open surgery started one hour and thirty minutes after diagnosis was revealed. During surgical exploration, the Ladd's bands were noted, as well as prevertebral duodenum and mobile colon. A long-standing intussusception was identified in ascending colon (Figure 4). The part of terminal ileum, cecum and part of ascending colon formed the intussusceptum. Ladd's bands were resected. The manual reduction of intussusception was impossible, because the bowel was dilated with ischemic and gangrenous wall layers. The resection of intussusceptum was performed. The total length of the resected intestine was 16cm. The end-to-end anastomosis of the rest of terminal ileum and transverse colon was performed and mesentery was reconstructed. At the end, standard Ladd's procedure was completed with the placement of the small intestine loops on the right and large intestine on the left. The procedure was successful eventually and lasted for 1h and 45 minutes. There were no postoperative complications. The infant was kept fasting for three days, with NG tube and antibiotic therapy (III generation cephalosporins and metronidazole). Intestinal passage normalized three days after the operation. Feeding was started on the fourth post-operative day. The recovery was successful, and the infant was discharged from hospital seven days after the procedure.

On the regular follow up one week after discharge, stitches were removed, wound was clear and healed. The infant had regular food intake as well as stool passage. After one month the infant was healthy, and follow-up terminated.

DISCUSSION

The age of WS presentation is variable and ranges from 13 days to 17 years in children. Although it can affect children at any age, it mainly affects children less than 2 years of age, with only 10–25% of cases occurring in older children [4]. One case has been recently noted in the adult population, in a 56-year-old patient [7]. Our patient was an infant, 5 months old, who presented with an acute form of this condition. Chronic intussusception is rare and accounts for about 3% of intussusceptions in children younger than one year and less than 10% in older children [4, 8]. Association of chronic intussusception with malrotation has only been described once in a literature [8, 9].

According to previously reported studies, which suggest that malrotation with mobile caecum, ascending and descending colon, increases the risk of ileocecal intussusception, often with long intestinal segments affected, that occurred in our patient [8]. Intussusceptum in our case was an ileocecal segment with a part of an ascending colon.

Our patient was referred with complaints about vomiting and abdominal pain with bloody mucus in a stool, without constipation or diarrhea. There were no abdominal distension and fever. It is in line with previously reported studies, but it is necessary to know that there are no absolutely certain signs for this association, and clinical presentation is always a combination of etiology as well as the time passed from the beginning of the symptoms [10, 11].

In our case, the diagnosis was based on ultrasound. At the beginning, the target sign was seen clearly, but afterwards the “whirlpool sign” and inverted ratio of superior mesentery artery and vein were noticed. Intestinal malrotation was confirmed by proximal gastrointestinal contrast study and stop in passage of contrast caudally. In some reported cases a CT scan was required to confirm the diagnosis [3, 11].

The most commonly used treatment methods for intestinal malrotation and intussusception are open manual reduction of intussusception and Ladd's procedure for malrotation. Laparoscopic approach is very rare [4]. Some reports indicate that many cases remain undetected, due to nonsurgical reduction of the intussusception with air or liquid enemas alone [5]. We did not try further hydrostatic reduction of intussusception due to ultrasound findings of both long intussusceptum and signs of malrotation, as well as the fact that almost 30 hours had passed since the beginning of symptoms. One study reported that intussusception manifested as anal prolapse because the entire colon was involved [5]. In our case, terminal ileum, cecum and ascending colon were affected, already gangrenous.

Our method of therapy was bowel resection and Ladd's procedure, because the attempt of manual reduction was not successful. In most of the reported cases intestine was viable and manual reduction was possible, but in some cases, resection was unavoidable [1, 3, 5, 8, 11, 14]. The postoperative course was uneventful in most of the reported cases, as well as in our case [1, 3, 4, 8].

In conclusion, in every pediatric patient with clinical and radiographic findings for intussusception, which does not respond to attempts of hydrostatic reduction, possible association of malrotation has to be considered and which must be sought after in a targeted manner. Time is essential factor, so do not hesitate to make a decision about surgery because it can prevent extended bowel resection.

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Conflict of interest: None declared.

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Figure 1. Upright native abdominal X ray on admission

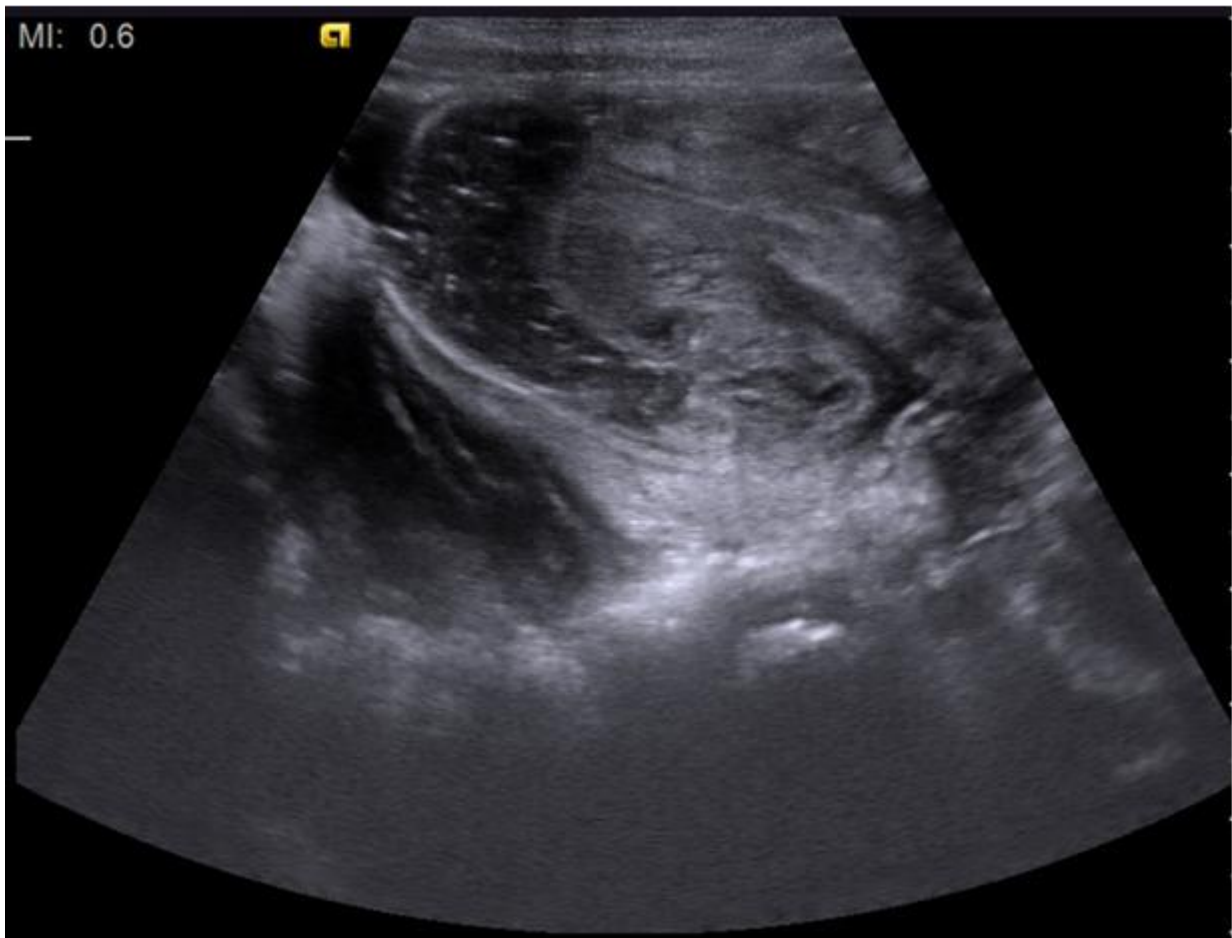


Figure 2. Abdominal ultrasound showing intestinal intussusception



Figure 3. Abdominal aultrasound showing „whirlpool sign“



Figure 4. Intraoperative view on intussusception