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Intussusception presenting with fluctuating mental status changes

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ABSTRACT

We report a 5 months old boy who was admitted to the PICU because of fluctuating level of consciousness. An extensive workup in search for infectious, neurologic, toxicologic and metabolic etiologies was done. Although abdominal symptoms were absent, he eventually was diagnosed with intussusception which needed surgery to recover. Intussusception should be included in the differential diagnosis of infants presenting with unexplained neurologic symptoms, ensuring timely diagnosis, treatment, and improved outcome.

1. Introduction

Encephalopathy in children could be a diagnostic challenge because of an often non-specific presentation and wide differential diagnosis. Encephalopathy is not a diagnosis, but a descriptive term for a syndrome of global brain dysfunction. Prompt recognition of the encephalopathic state and appropriate investigation and treatment help to reduce morbidity and mortality [1,2].

2. Case presentation

A previously healthy 5 months old boy was admitted to the PICU because of fluctuating level of consciousness, which is a sign of encephalopathy. The first sign of illness occurred in the morning when he suddenly arched his back and stretched backwards. Later that day, he vomited once, and hereafter he had been intermittently crying and was less responsive to his parents. He did not have a fever, seizures, or a trauma prior to admission, and there was no history of intoxication. The past few days, he had a runny nose and stools were thinner than usual. A developmental delay was not present.

Vital signs showed a respiratory rate of 40 breaths/minute, 100% oxygen saturation, heart rate 140 beats/minute, capillary refill time 2 s, blood pressure 90/60 mmHg, and temperature 36.4 °C. Neurological examination revealed an altered mental status; periods of somnolence (i.e. diminished response to parents) and pinpoint pupils interchanged with periods of adequate behaviour and normal pupil reactions to light. Motor function and palmar grasp reflexes were normal. Meningeal irritation was absent. There were no signs of dehydration. Abdominal examination showed normal peristalsis, no tenderness, no abdominal masses, and no hepatomegaly. Further physical examination was unremarkable.

Laboratory investigation showed white blood cells 23x10^9/L, CRP <1 mg/L, glucose 9.7 mmol/L, sodium 138 mmol/L, potassium 3.8 mmol/L, calcium 2.62 mmol/L, magnesium 0.75 mmol/L, phosphate 1.73 mmol/L, blood urea nitrogen 2.5 mmol/L, creatinine 20 μmol/L, ammonia 57 μmol/L, pH 7.41, pCO2 35 mmHg, base-excess −2 mmol/L, and bicarbonate 22 mmol/L. Imaging of the brain by ultrasound and MRI with contrast revealed no abnormalities. Cerebrospinal fluid (CSF) analysis was performed after imaging of the brain and revealed white blood cells 2x10^6/L, protein 0.27 g/L, and CSF-to-serum glucose ratio 0.5. Also, an electroencephalogram showed no abnormalities. Toxicology screening in urine was negative. Plasma and urine were stored to screen for inborn errors of metabolism.

The initial working diagnosis was meningitis/encephalitis, mainly based on the presenting symptoms and physical examination. Antibiotic (Ceftriaxone, IV administration, daily dosage of 100 mg/kg) and antiviral (Acyclovir, IV administration, daily dosage of 1500mg/m2, dosage every 8 h) therapy was prescribed awaiting microbiology and virology results. Other central nervous system possibilities of encephalopathy, such as trauma, hemorrhage, or abscess were ruled out because of normal imaging. Epileptic activity was unlikely because of a normal electroencephalogram. Also, there were no signs of intoxication or

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electrolytes imbalances. At this stage, intussusception was not included in the differential diagnosis, but should have been considered in an infant with unexplained neurologic symptoms.

Our patient gradually became more comfortable and alert, and although he developed a high blood pressure of 120/60 mmHg, he was discharged from the PICU to the general pediatric ward because his neurologic condition had been improved. However, approximately 48 h after PICU admission, an abdominal X-ray and ultrasound was done because of somnolence and bilious vomiting. The abdominal ultrasound revealed a target-sign suspected for intussusception(Fig. 1).

After diagnosis of intussusception, reduction by hydrostatic enema was unsuccessful, thus the ileocaecal intussusception was repaired surgically by end-to-end anastomosis.

Microbiologic testing eventually showed negative bacterial results for CSF and blood. No viral infection was found in CSF and blood. Antibiotic and antiviral therapy were terminated 48 h after CSF and blood samples were taken. A throat swab was positive for parainfluenza virus and stool was positive for adenovirus.

The immediate day after surgery, our patient did not have any complaints; he was alert, active, and the blood pressure restored from hypertension to normal values. He was discharged home four days after admission to the hospital.

3. Discussion

Neurologic symptoms as presenting symptoms of intussusception, for example changes in consciousness level, lethargy, or irritability, are not rare and have been described in up to 70% of children with intussusception [3–5]. The presence of neurologic symptoms is associated with younger age and with a more sudden onset of disease symptoms [3]. These neurologic symptoms often accompany classic symptoms of intussusception such as vomiting, abdominal pain, and passage of blood per rectum. However, occasionally only neurologic symptoms are present which may lead the clinician to a diagnosis of intracranial pathology and may result in an extensive workup in search for infectious, neurologic, toxicologic and metabolic etiologies [6]. The resulting delay to diagnose intussusception is important to underscore, since delay to diagnosis decreases the success of radiologic reduction and increases the risk of operative intervention and bowel resection [7].

The pathophysiology of neurologic manifestations of intussusception is not clearly understood. One hypothesis is that the body produces endogenous opioids to reduce abdominal pain. This theory is based on case-reports of children with intussusception presenting with pin-point pupils and altered consciousness who recovered after naloxone [8]. However, a study in 8 children with barium enema proven intussusception (of which 2 presented with lethargy) showed that plasma beta-endorphin level were not higher than in children without intussusception [9]. Another hypothesis is that intussusception causes bowel ischemia which releases toxic metabolites and uncharacterized cytokines which are known to affect the central nervous system causing lethargy or encephalopathy. These substances may achieve higher concentrations in the immature brain of children, and could possibly explain why neurologic symptoms occur more often in younger children [10,11].

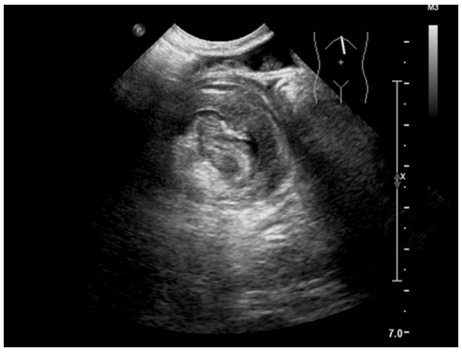

Fig. 1. Abdominal ultrasound showing a target-sign suspected for intussusception.

In conclusion, we report a potential pitfall for clinicians. Changes in level of consciousness could be the presenting symptom of intussusception, a common problem in children, even in the absence of abdominal symptoms. Intussusception should be included in the differential diagnosis of infants presenting with unexplained neurologic symptoms and could lead to early diagnosis and treatment, and subsequently improve patient outcome. Endogenous opioid poisoning is hypothesized to cause miosis and encephalopathy. Further studies are needed to clarify the pathophysiology of neurological manifestations in acute abdominal disorders.

3.1. Patient perspective

"I [mother] was really scared, I thought my son was about to die when I saw him alternating between crying and seemingly loss of consciousness. I picked him up and ran to seek for medical help. After extensive evaluation at the Emergency Department, the initial thoughts were that he had meningitis/encephalitis. We still were afraid that we were going to lose our son to this serious condition. In our communication with the medical team, the team explained that not all results totally fit the diagnosis of meningitis/encephalitis and the team therefore was not sure if the diagnosis was correct. We really appreciated the honesty of the team at that point. Because fluctuating level of consciousness and pinpoint pupils persisted, we were transferred to the PICU. Nevertheless, after the first dosage of antibiotics, our son became more alert and active and we felt relieved that he was getting better. He subsequently was discharged to the general pediatric ward, which strengthened our feeling that he was getting better. However, something felt wrong. Although he was more alert and finished his bottles, he was sleeping a lot of the time and he was vomiting. When bilious vomiting was noticed, an abdominal ultrasound was done diagnosing intussusception. After surgery, he soon got better and restored to normal.

Despite the initial concerns, we have fond memories of the hospital stay: we felt taken seriously and we were updated regularly in understandable language. Nowadays, three years after admission to PICU, our son does not have any complaints of abdominal pain. He recently discovered his scar and finds this an interesting part of his body. Psychologically, he initially seemed to be scared for elevators (we took a lot of elevators during our hospital stay), but as the years passed by, this problem disappeared. He does not have any problems at the moment; his growth and development is according to his age and he is a young, active, and happy boy."

Patient consent

Both parents have read the manuscript and have given written informed consent to publish this case report.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence

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