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Case Report

Abdominal Epilepsy, A Diagnosis to Consider in Children with Recurrent Vomiting: A Case Report

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ABSTRACT

Introduction: Abdominal epilepsy is a rare disorder characterized by recurrent gastrointestinal complaints and central nervous system involvement, mainly confusion, fatigue, headache, dizziness, syncope, and lethargy resulting during the abdominal seizure activity. Chronic vomitting or other gastrointestinal symptoms often lead to fluid and nutritional deficiencies which resulted in malnutrition. To date, there was no study has been reported on abdominal epilepsy in Indonesia.

Case Presentation: We presented a case of two-year-old girl with chief complaint of recurrent profuse vomiting every month for the past year. Patient complained of headaches and abdominal pain and tended to sleep all day during relapse. Physical examination showed sunken eyes and decreased skin turgor. EEG showed abnormal results, diffuse irritative, epileptiform with normal basic rhythm. Esophageal polyps were also found on endoscopy. Patient was given anti-epileptic drugs and histamine H_2 receptor antagonists and responded well to those therapies.

Conclusion: Abdominal epilepsy is a rare case which may resulted in vomiting and abdominal pain in children. EEG examination is an important test for diagnosing abdominal epilepsy. Treatment with anti-epileptic drugs is known to respond well in this case.

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INTRODUCTION

Abdominal epilepsy is a type of epilepsy. This disease is more often as a diagnosis of exclusion. Abdominal epilepsy is reported to be more in pediatric patients than adults [1]. It is also reported more common in women, with majority of 9–12 years old [2]. According to a study in Diyala Province, Iraq, abdominal epilepsy were found in 34 patients (7.5%) out of 450 patients, mostly between 6 to 12 years old [3].

Abdominal epilepsy resulted in gastrointestinal symptoms. Some gastrointestinal symptoms that occurred in patients with abdominal epilepsy are abdominal pain, vomiting, nausea, bloating, and diarrhea. These symptoms recurred abruptly with no explanation [4]

Diagnosis of abdominal epilepsy is established in a child with recurrent gastrointestinal symptoms, symptoms of central nervous system disorders (such as confusion, fatigue, headache, and fainting), abnormalities on electroencephalography (EEG), and respond well to antiepileptic therapy [4]. There was no study has been reported on abdominal epilepsy in Indonesia. Clinicians in primary health services rarely consider the possibility of abdominal epilepsy in children with recurrent and chronic vomiting due to scarcity of studies. Early diagnosis may prevent fluid and nutritional deficiencies which resulted in malnutrition due to repeated chronic vomiting.

CASE PRESENTATION

A two-year-old girl was presented to the hospital by her family with complaint of vomiting in the last 3 days, up to 20 times a day, yellow in color, containing what the patient has eaten and drunk. Patient also

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experienced headache and abdominal pain with no diarrhea and no fever. Patient had difficulty with eating, weakness, tended to sleep throughout the day, had not urinated for more than 6 hours and had lost 4 kilograms. The patient has a history of epilepsy since he was 13 months old. Antiemetic drugs were administered by a local midwife, but there was no response.

Parents took the patient to a pediatrician, who suspected gastritis, antiemetic drugs and histamine H2 receptor antagonist drugs did not relieve the complaints. The patient was hospitalized for several days with similar complaints (vomiting) and dehydration. Patient complained of recurrent vomiting for the past six months, 4–7 days every month, more than 20 times per day. Patient complained of headache, abdominal pain, and tended to sleep throughout the day in each period of relapses. Physical examination showed sunken eyes and decreased skin turgor.

Medical Family History

In the family, the younger brother of her grandfather was known to have a history of seizures but currently had never recurred.

Obstetric History

The patient's delivery were assisted by midwife, full term, weighing 3400 grams. During pregnancy, her mother had experienced common cold for 2 weeks which resulted in her mother being hospitalized due to fatigue.

Psychosocial History

The patient is the youngest child of two who is living with her parents and grandparents. She has Javanese ethnic family. She is not currently attending a school so her mother teaches her to color, memorizes animal names and learns english at home.

Neurological Examination

After six months, the vomiting continued to occur so an electroencephalography examination was performed with abnormal results: diffuse irritative, epileptiform with normal basic rhythm.

Other Examinations

Laboratory results showed leucopenia ($2{,}410^3/\mu L$) and mild hyponatremia (134.0 mmol/L). Endoscopic examination was also performed to trace the causes of recurrent vomiting and other possible etiologies. Endoscopy revealed a single polyp in distal esophagus and mild gastroduodenitis.

Diagnosis

After considering main symptoms and results from examinations such as complete blood test, electroencephalography and endoscopic examination, diagnosis of esophageal polyps, mild gastroduodenitis, chronic vomiting with moderate mild dehydration, abdominal epilepsy, and underweight were established and the patient was treated by an attending physician.

Treatment and Follow-up

Rehydration and 1.25 cc per 12 hours of valproic acid (14 mg/kilogram body weight/day) were administered after EEG result, but two weeks later, patient experienced repeated vomiting for three days more than 20 times a day. Dose of valproic acid was increased to 1.35 cc per 12 hours (15 mg/kilogram body weight/day). One week later, patient still reported vomiting for about 3 days, 8 to 10 times a day, thus valproic acid doses increased to 1.5 cc per 12 hours.

Valproic acid doses were increased to 1.6 cc per 12 hours and ranitidine 17 mg per 12 hours were administered to the patient after endoscopy result. Two weeks later, patient reported recurrent vomiting for 2

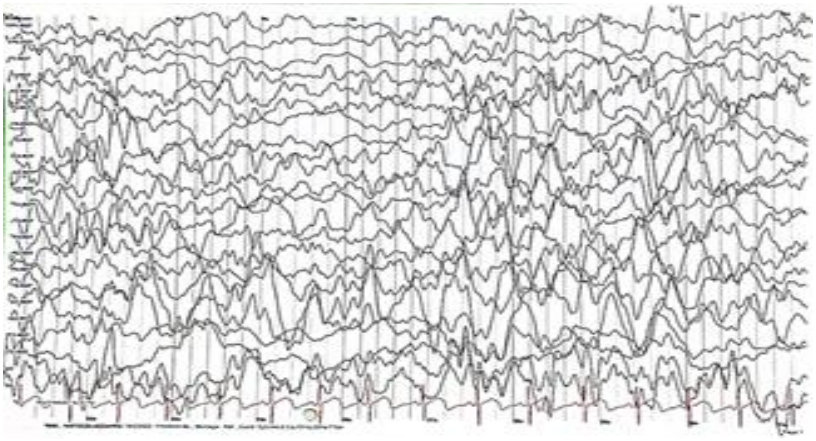


Fig 1. Electroencephalography Result. Abnormal results, diffuse irritative epileptiform with normal basic rhythm

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Fig. 2. The Endoscopic Result. (A) Showing friable polypoid tissue in the LES, (B) Hyperemic gastric corpus mucosa, (C) Mild hyperemic duodenal bulbus

days, more than 8 times a day. Valproate acid doses were increased to 2 cc per 12 hours. Recurrence only occurred once a month with duration of 3 days and frequency of vomiting was only 1 to 4 times per day with the dose.

DISCUSSION

Abdominal epilepsy is a rare disorder characterized by recurrent gastrointestinal complaints resulting from seizure activity. These symptoms are supported by abnormal electroencephalography (EEG) results [5]. Currently, use of abdominal epilepsy term is still controversial among neurologists. Tatum (2020) recommended changing the term of abdominal epilepsy to focal aware motor seizures with abdominal clonic jerking because this term may explain this disease better [6].

Incidence rate of abdominal epilepsy is not clearly known. Zinkin & Peppercorn (2004) stated in their study that there were 36 cases of abdominal epilepsy in UK. In India, 74% of 150 children with recurrent abdominal pain and other gastrointestinal symptoms had abnormal EEG results. EEG abnormalities were found in children of 6–15 years old. Abdominal epilepsy was found in 67.56% females and 32.43% males. EEG abnormalities in patients with recurrent gastrointestinal symptoms are one of the criteria for diagnosing abdominal epilepsy [2].

One hypothesis explained mechanism of abdominal epilepsy is due to presence of abnormal waves in temporal lobe and amygdala area. Through the dorsal motor nucleus of vagus nerve, amygdala transmits neurotransmission to gastrointestinal tract. The amygdala is also thought to transmit transmission to the hypothalamus, then hypothalamus activate the sympathetic pathways, and cause gastrointestinal symptoms [7].

There are four criteria for diagnosis of abdominal epilepsy, [a] gastrointestinal symptoms such as abdominal pain often occurs in periumbilical or upper abdominal region nausea, recurrent diarrhea and vomiting, or a combination [4,7,8]. Frequency of vomiting is usually 3 attacks in 1 hour to 10 days in the last 6 months. In each attack, patient can vomit four times in one hour [9]. [b] Central nervous system symptoms include confusion, fatigue, headache, dizziness, syncope, and lethargy [4]. Disorientation during the episode followed by fatigue and tendency to sleep are important symptoms to establish the diagnosis [10]. [c] Electroencephalography is an important and primary examination in diagnosing abdominal epilepsy [11]. Patients with abdominal epilepsy have specific abnormalities on EEG. Most often, EEG showed temporal lobe seizures, generalized spikes, wave discharges, and frontal lobe discharges [12]. In a study of Kshirsagar *et al*. (2012), showed that of 111 abnormal EEGs in patients with recurrent gastrointestinal symptoms, 35.13% of the abnormal waves were in temporal lobe, 32.43% in the frontotemporal lobe, and 29.72% had generalized type EEG results, with 89.18% of the wave pattern in the form of sharp waves. Examination of blood, cerebrospinal fluid, abdominal ultrasound, gastrointestinal computed tomography (CT) scan, and gastrointestinal endoscopy in abdominal epilepsy is usually normal [9]. [d] Patients with abdominal epilepsy are known to improved with antiepileptic drugs. Common antiepileptic drugs administered to patients with abdominal epilepsy in children are phenobarbital, valproic acid, and carbamazepine [13,14].

Paroxysmal gastrointestinal symptoms are symptoms that are not only found in abdominal epilepsy but also can be found in porphyria, familial Mediterranean fever, cyclical vomiting syndrome and abdominal migraine. Most common difficulties experienced by clinicians is to differentiate between abdominal epilepsy and abdominal migraine, in terms of symptoms. Duration of symptoms and EEG results may be used to differentiate the two entities. EEG in patients with abdominal epilepsy usually shows specific

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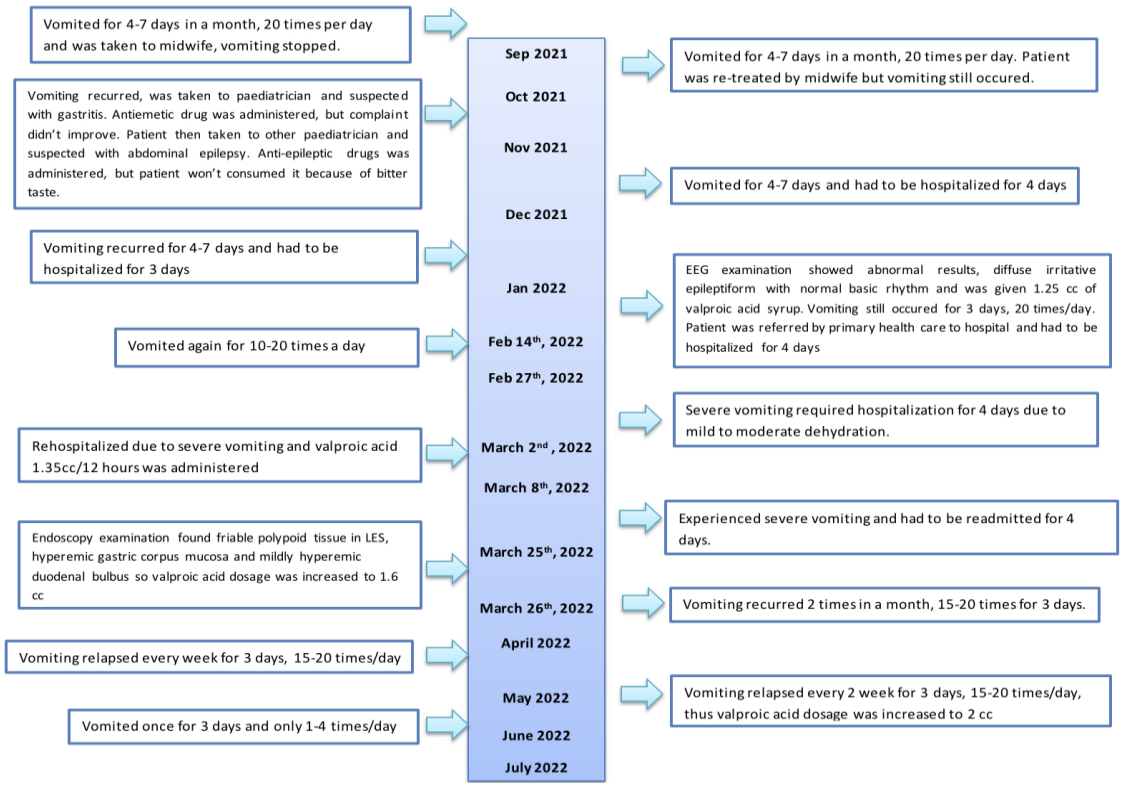


Fig. 3. Chronologies of The Case

abnormalities, most common abnormalities are found in the temporal lobe. Meanwhile, in abdominal migraine, the duration of complaints is longer and no abnormalities were found in the EEG results [1,15].

Our patient met all the diagnostic criteria for abdominal epilepsy. Gastrointestinal complaints of our patient are recurrent profuse vomiting and abdominal pain that has recurred every month. Central nervous system symptoms in our patient are headaches and tend to sleep all day during relapse. The presence of abnormalities in EEG results of our patient is diffuse, irritative, epileptiform with normal basic rhythm. We also found a single polyp in distal esophagus and mild gastroduodenitis after endoscopic examination. It can be formed as a result of repeated vomiting. These polyps grow from an exaggerated response to mucosal injury due to infection, surgical injury, or reflux of gastric fluid that comes out during vomiting process [16].

Valproic acid was administered to our patient. Prior to administration of valproic acid, complaints appeared 4 to 7 days per month, with the frequency of vomiting more than 20 times a day. After administration of valproic acid and multiple dosage increases, intensity and frequency of complaints decreased. Every month, complaints only appear for 3 days and the frequency of vomiting and abdominal pain decreased to 1 to 4 times a day. This showed that our patient has a good response to anti-epileptic drugs. Frequency of vomiting was significantly reduced and no neurological symptoms were found after 5 months of administration of valproic acid. This is in accordance with a study conducted by Cerminara *et al.* (2013) that revealed 15 of 18 patients with abdominal epilepsy responded well to antiepileptic drugs. Twelve cases no longer showed abdominal epileptic symptoms after treatment. Common anticonvulsants or antiepileptic for pediatric patients with abdominal epilepsy are carbamazepine, valproic acid, phenobarbital, oxcarbazepine, and phenytoin [17]. A study by Ostwal in 2013 showed that 88% of patients improved after administration of oxcarbazepine.

CONCLUSION

Abdominal epilepsy is a rare case which may result in recurrent vomiting and abdominal pain in children especially with a history of epilepsy. Gastrointestinal and neurological symptoms should be questioned more specifically by attending physician. EEG examination is an important test for diagnosing abdominal epilepsy. Treatment with anti-epileptic drugs is known to respond well in this case. In the future, abdominal epilepsy can be considered as a differential diagnosis in children with recurrent vomiting after exclusions of possible life-threatening etiologies of abdominal pain and recurrent vomiting in children, thus proper examination and treatment can be carried out earlier.

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CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

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