DOI: 10.7860/JCDR/2024/67269.19207

Case Series

Paediatrics Section

Unusual Presentations of Epilepsy: A Series of Paediatric Cases with Abdominal Epilepsy and Epileptic Angina

RITA PANYANG KATAKI[1], BHASKAR BORAH SONOWAL[2], KOMAL AGARWAL[3], CHOW NIFASENG CHIRING[4], LUHIT DHUNGEL[5]

ABSTRACT

Epilepsy, a common neurological disorder characterised by recurrent seizures, can manifest in unusual clinical presentations such as abdominal pain, nausea, vomiting, and bloating associated with diverse Central Nervous System (CNS) symptoms like confusion, fatigue, headache, dizziness, and syncope. The importance of recognising and documenting such unusual presentations lies in their potential to be misdiagnosed or overlooked, delaying appropriate treatment and causing undue distress to both the patients and their families. In the present case series, patients presented with gastrointestinal or chest pain for a prolonged period. As the symptoms did not subside with treatment, an Electroencephalogram (EEG) was performed to rule out epilepsy. In all three cases (10-year and 10-month-old female, 11-year-old male, seven-year-old male), EEG revealed a generalised seizure disorder. The present case series highlights unusual presentations of epilepsy in paediatric patients, including Abdominal Epilepsy (AE) and epileptic angina. These cases underscore the importance of considering epilepsy as a potential aetiology in patients with recurrent and atypical symptoms.

Keywords: Cyclical vomiting, Electroencephalogram, Seizure disorder

INTRODUCTION

Epilepsy, a common neurological disorder characterised by recurrent seizures, can manifest in a diverse range of clinical presentations. While the classical manifestations of epilepsy include generalised or focal motor seizures, some cases present with atypical and puzzling symptoms. In paediatric populations, the recognition and accurate diagnosis of uncommon seizure presentations become even more challenging due to the limited ability of young children to articulate their symptoms effectively [1].

The authors presented three unusual presentations of epilepsy in the present case series. Through the present case series, the authors aimed to shed light on the clinical features, diagnostic challenges, and management strategies for AE and epileptic angina in paediatric patients. By sharing the experiences with these unique cases, the authors hope to contribute to the growing body of knowledge that supports the timely recognition and optimal management of rare epilepsy presentations in the paediatric population.

Case 1

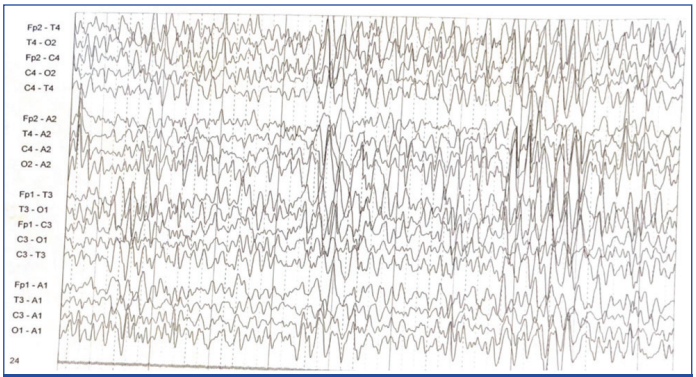
A 10-year and 10-month-old female patient was admitted to the Department of Paediatrics with a complaint of repeated episodes of vomiting associated with intense feelings of uneasiness and weakness for the last two days. The patient has been experiencing similar symptoms since the age of 4.5 years. Since then, she has had recurrent episodes of vomiting lasting for a few hours without any abdominal pain or fever, leading to numerous hospital admissions every 2-3 months over the last six years. There is no family history of a similar disease.

Physical examination, including abdominal and neurological examinations, was unremarkable. During each episode, the patient was managed as a case of acute gastritis, receiving conservative treatment including intravenous fluids such as Ringer's lactate and normal saline, as well as the proton pump inhibitor pantoprazole (1 mg/kg) and the antiemetic drug ondansetron (0.1 mg/kg).

Based on the history provided, the differential diagnosis included acute gastritis, abdominal migraine, psychogenic vomiting, intracranial space-occupying lesion, and AE.

The patient underwent a comprehensive battery of investigations, including a complete blood count, serum electrolytes, liver function tests, renal function tests, and imaging studies such as abdominal ultrasonography and a barium meal. All blood investigations were within normal limits. Additionally, a Non Contrast Enhanced Computed Tomography (NECT) scan of the brain was performed to rule out central nervous system abnormalities. The NECT brain revealed no abnormalities, and the patient appeared normal between episodes.

Given the recurrent episodes of cyclical vomiting and the absence of any organic pathology in the investigations, AE was suspected as a differential diagnosis. To confirm this, an EEG was recommended. The EEG exhibited characteristic wave patterns suggestive of a seizure disorder, confirming the diagnosis of AE [Table/Fig-1a].



[Table/Fig-1a]: EEG finding of generalised seizure disorder (Case 1).

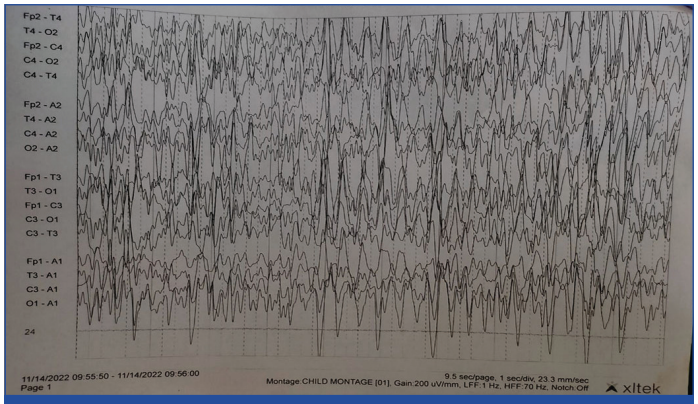
Upon confirmation of the diagnosis, the patient was initiated on Valproic acid 20 mg/kg/day in two divided doses and advised to continue until further advised, atleast up to two years. Subsequent follow-up visits every month demonstrated a favourable response to treatment, with the patient remaining asymptomatic until the last follow-up, one year after discharge.

Case 2

An 11-year-old male child presented with a complaint of frequent left-sided chest pain for almost two months and was admitted to

the Paediatric Ward of Assam Medical College and Hospital. He had a history of prior hospitalisation for a similar complaint one month back in the hospital. The chest pain was described as sharp and excruciating, with breathing difficulty. There were no alleviating or relieving factors of pain, it was localised in nature, and occurred at any time of the day, lasting for 5 to 10 minutes and subsiding by itself due to tiredness. There were no episodes of loss of consciousness or abnormal movements. There was no family history of a similar disease.

There were no significant findings in general and systemic examinations. Extensive cardiovascular investigations, including an Echocardiogram (ECG), and chest radiograph, were performed to assess the patient's cardiovascular system. All these investigations yielded normal results, ruling out cardiac pathology, and the condition was treated as acute gastritis. The patient was treated with injection ondansetron (0.1 mg/kg) and injection ranitidine (2 mg/kg), which failed to provide relief from the symptoms. The patient's neurological examination, as well as his vital signs, were within normal limits. Blood investigations, including renal function and electrolyte assessment, showed no abnormalities. Given the recurrent nature of the symptoms, the possibility of myoclonic seizure, atonic/tonic, or absence seizure was considered. An EEG was performed, which revealed generalised high voltage sharp and slow waves, consistent with a diagnosis of generalised seizure disorder [Table/Fig-1b].

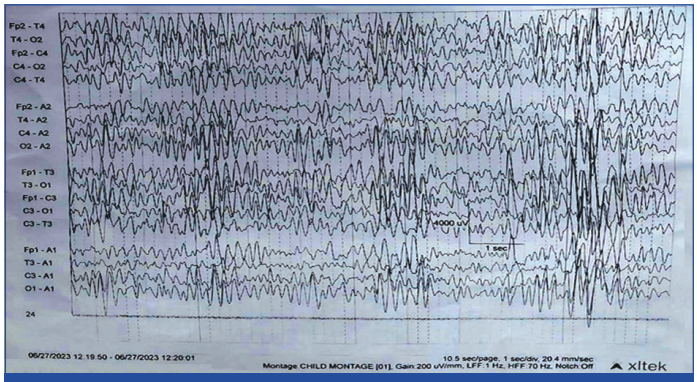


[Table/Fig-1b]: EEG finding of generalised seizure disorder (Case 2).

requiring three hospital admissions within one year. Each time, the patient was treated for acute gastritis and discharged. General and local examination of the abdomen revealed no abnormalities. Initial treatment with a proton pump inhibitor, pantoprazole (1 mg/kg), paracetamol (15 mg/kg), and ondansetron (0.1 mg/kg), started in the hospital, failed to provide relief from the symptoms. Complete blood picture results were normal, ruling out systemic infection or inflammation. Given the recurrent and atypical presentation of the symptoms, the possibility of an epileptic aetiology was suspected. Other differential diagnosis were acute gastritis and abdominal migraine. An EEG was performed, which revealed abnormal findings in the form of generalised high voltage sharp and slow wave activity, consistent with a diagnosis of generalised seizure disorder [Table/Fig-1c]. Based on the EEG findings and the diagnosis of generalised seizure disorder, the patient was started on sodium valproate at 20 mg/kg/day. The patient was followed-up for six months at regular intervals of four weeks and remained symptom-free during follow-up. Clinical findings of all three cases are described in [Table/Fig-2].

DISCUSSION

Abdominal Epilepsy (AE) is a rare form of epilepsy where seizures manifest primarily with abdominal symptoms, such as abdominal pain, nausea, vomiting, or gastrointestinal disturbances, potentially leading to misdiagnosis as gastrointestinal disorders. EEG abnormalities and positive responses to the introduction of Antiepileptic Drugs (AED)



[Table/Fig-1c]: EEG findings of generalised seizure disorder (Case 3).

Case	Age/gender	Symptoms	EEG	Diagnosis	Treatment received	Follow-up
1	10 y 10 m/F	Paroxysmal cyclical vomiting associated with intense feelings of anxiousness for six years	Generalised high voltage sharp and slow wave activity	Abdominal epilepsy	Sodium valproate @20 mg/kg/day in two divided dose	At 12 months, symptom-free
2	11 y/M	Sharp and excruciating left-sided chest pain for two months	Generalised high voltage sharp and slow waves activity	Epileptic angina	Sodium valproate @20 mg/kg/day in two divided dose	At 6 months, symptom-free
3	7 y/M	Non projectile vomiting and acute epigastric pain for one year	Generalised high voltage sharp and slow wave activity	Abdominal epilepsy	Sodium Valproate @20 mg/kg/day in two divided dose	At six months, symptom-free

[Table/Fig-2]: Summary of all the three cases in present study.

The patient was initiated on sodium valproate at 20 mg/kg/day in two divided doses. Following the commencement of treatment, the patient's symptoms completely resolved within one week, and the patient was successfully discharged. The patient was followed-up every four weeks for the last six months, and he was symptom-free during the follow-up.

Case 3

A seven-year-old male presented to the paediatric Outpatient Department (OPD) with multiple episodes of non projectile vomiting and acute abdominal pain over the past two days. Each vomiting episode was preceded by epigastric pain and lethargy. The abdominal pain was localised mainly in the epigastric region and worsened after food intake. The pain was sharp, aching, continuous in nature, lasting for 5-10 minutes, and occurring several times a day. The patient also developed low-grade fever during the episodes. Notably, the child had a history of similar complaints in the past with acute abdominal pain followed by nausea and vomiting,

are common [2]. Conversely, epileptic angina, also known as ictal angina, is characterised by chest pain and discomfort during seizures, mimicking cardiac ischaemia, which can result in unnecessary cardiac investigations and interventions [3]. Recognising and documenting such unusual presentations is important due to the potential for misdiagnosis or oversight, which could delay appropriate treatment and cause undue distress to both the patients and their families. Early and accurate diagnosis is crucial for the prognosis and management of epilepsy, underscoring the importance of increasing awareness among healthcare professionals about these uncommon seizure presentations [4].

The recurrent episodes of cyclical vomiting and abdominal pain in these cases led to multiple hospital admissions and exhaustive investigations to rule out organic pathology. However, in the absence of any abnormalities in the investigations and the observation of normalcy between episodes, suspicion of an epileptic aetiology arose [4]. A comparison of the findings in the present cases is made with the findings of previous studies in [Table/Fig-3] [5-8].

Name of the author	Place/Year	Presentation	Treatment
Mondal R et al., [5]	Siliguri, West Bengal/2014	Case series of Abdominal epilepsy in 6 patients who presented with recurrent vomiting and pain.	Treated with antiepileptic and responded well. Two patients were given phenytoin, 2 were given carbamazepine and 2 were given valproic acid.
Sureshbabu S et al., [7]	New Delhi/2017	Epileptic angina on a 14 year boy presented with localised chest pain.	Started with 100 mg of zonisamide and switch to 400 mg of carbamazepine and symptom-free on four months follow-up.
Dutta SR et al., [6]	Guwahati, Assam/2007	Case series of abdominal epilepsy in three patient, two paediatrics and one adult recurrent abdominal pain.	All the three case were treated with Oxcarbazepine and symtom-free on follow-up.
Singhi PD and Kaur S et al., [8]	Chandigarh/ 1988	Case of 10-year-old male, Abdominal epilepsy presenting with recurrent paroxysmal abdominal pain.	Initially phenobarbitone, then phenytoin. Patient was symptom-free on six months folow-up.
Present study	Dibrugarh , Assam/2023	Cases series of 3, 2 abdominal epilepsy and one epileptic angina.	All three cases responded well to valproic acid. Patients were symptom-free on follow-up.

[Table/Fig-3]: Comparison of findings of present study with previous studies [5-8].

Mondal R et al., from Siliguri compiled a case series of AE in which six patients were diagnosed with AE, noting that, with the exception of two patients who experienced recurrent vomiting, recurrent pain was a prevalent occurrence among all the other patients [5]. Dutta SR et al., from Assam published a case report of three AE cases, one of which involved an adult. All patients responded well to antiepileptic medication [6]. Sureshbabu S et al., shed light on a similar case report of a 14-year-old boy who exhibited recurring, brief, localised chest pain. Cardiac and systemic assessments were unremarkable, and brain Magnetic Resonance Imaging (MRI) showed no structural issues. Video telemetry was performed to understand the episodes. The boy was diagnosed with epileptic angina based on EEG findings. A predominantly centroparietal rhythm was observed in the left hemisphere during seizures. The patient responded notably well to antiseizure medications [7]. Singhi PD and Kaur S reported a 10-year-old male child from Chandigarh with recurrent paroxysmal abdominal pain initially treated as psychogenic pain, later diagnosed as AE, based on EEG findings, and responded well to antiepileptic treatment [8].

In the present case series, EEG played a crucial role in confirming the diagnosis of epilepsy. The characteristic wave patterns observed in the EEG were consistent with a seizure disorder, leading to the diagnosis of AE. Initiation of antiepileptic treatment, specifically sodium valproate, in these cases resulted in a favourable response, with both patients becoming asymptomatic and remaining symptom-free on follow-up.

In epileptic angina, the primary symptom is chest pain, often raising concerns for cardiac pathology [7]. Epileptic pain as a somatosensory manifestation of seizures is not uncommon, but as a solitary presentation of epilepsy is extremely rare and diagnostically challenging [7]. Pazarci NK et al., searched their database of 4736 patients and identified only nine patients with this epileptic pain symptom. They found symptoms such as nuchal pain, headache, abdominal pain, and pain in the limbs. The ictal EEG often showed hemispheric or diffuse abnormalities rather than focal, well-defined ictal activity [9]. In case 2, a previously healthy 11-year-old child presented with sharp, excruciating left-sided chest pain, accompanied by breathing difficulty lasting 5-10 minutes.

These cases emphasise the importance of maintaining a high index of suspicion for epilepsy in patients with recurrent and atypical symptoms, especially when standard investigations yield negative results. EEG is a valuable tool in diagnosing epilepsy and should be promptly considered when neurological involvement is suspected.

Notably, all three cases showed a favourable response to treatment with sodium valproate, highlighting its efficacy in managing epilepsy in paediatric patients with these unusual presentations. However, further research and larger studies are needed to better understand the prevalence, pathophysiology, and optimal management of AE and epileptic angina in the paediatric population.

CONCLUSION(S)

The present case series highlights unusual presentations of epilepsy in paediatric patients, including AE and epileptic angina. These cases underscore the importance of considering epilepsy as a potential aetiology in patients with recurrent and atypical symptoms. Prompt diagnosis and initiation of treatment with sodium valproate led to favourable outcomes, resolving symptoms and improving patient well-being.

REFERENCES

[1] Fisher RS, Acevedo C, Arzimanoglou A, Bogacz A. 'ILAE official report: A practical clinical definition of epilepsy'. Epilepsia. 2014;55(4):475-82. Doi: 10.1111/epi.12550. PMID 24730690. S2CID 35958237.
[2] Franzon RC, Lopes CF, Schmutzler KM, Moreis MI, Guerreiro MM. Recurrent abdominal pain: When an epileptic seizure should be suspected? Arquivos de Neuro-Psiquiatria. 2002;60(3-A):628-30.
[3] Shorvon S, Tomson T. Sudden unexpected death in epilepsy. The Lancet. 2011;378(9808):2028-38.
[4] Zinkin NT, Peppercorn MA. Abdominal epilepsy. Best Pract Res Clin Gastroenterol. 2005;19(2):263-74.
[5] Mondal R, Sarkar S, Bag T, Mondal K, Saren A. A paediatric case series of abdominal epilepsy. World J Pediatr. 2014;10(1):80-82.
[6] Dutta SR, Hazarika I, Chakravarty BP. Abdominal epilepsy, an uncommon cause of recurrent abdominal pain: A brief report. Gut. 2007;56(3):439-41.
[7] Sureshbabu S, Nayak D, Peter S, Sobhana C, Mittal G. Epileptic angina. Epilepsy & Behavior Case Reports. 2017;7:49-53.
[8] Singhi PD, Kaur S. Abdominal epilepsy misdiagnosed as psychogenic pain. Postgrad Med J. 1988;64(750):281-82.
[9] Pazarcı NK, Bebek N, Baykan B, Gürses C, Gökyiğit A. Reappraisal of epileptic pain as a rare symptom of seizures. Epilepsy Behav. 2016;55:101-07.

PARTICULARS OF CONTRIBUTORS:
1. Associate Professor, Department of Paediatrics, Assam Medical College, Dibrugarh, Assam, India.
2. Senior Resident, Department of Paediatrics, Assam Medical College, Dibrugarh, Assam, India.
3. Resident, Department of Paediatrics, Assam Medical College, Dibrugarh, Assam, India.
4. Senior Resident, Department of Paediatrics, Assam Medical College, Dibrugarh, Assam, India.
5. Senior Resident, Department of Paediatrics, Assam Medical College, Dibrugarh, Assam, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Rita Panyang Kataki,
House No. 28, West Milan Nagar, Lane I, Dibrugarh-786003, Assam, India.
E-mail: panyang.rita.dr@gmail.com

PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Sep 02, 2023
- Manual Googling: Nov 28, 2023
- iThenticate Software: Jan 27, 2024 (5%)

ETYMOLOGY: Author Origin

EMENDATIONS: 8

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? No
- For any images presented appropriate consent has been obtained from the subjects. No

Date of Submission: **Aug 30, 2023**
Date of Peer Review: **Nov 11, 2023**
Date of Acceptance: **Jan 31, 2024**
Date of Publishing: **Mar 01, 2024**