A pediatric case series of abdominal epilepsy

Rakesh Mondal, Sumantra Sarkar, Toshibananda Bag, Kalyanbrata Mondal, Abhisek Saren
Siliguri, India

Background: **Abdominal epilepsy (AE) is an infrequent cause of recurrent abdominal pain in children. It is characterized by paroxysmal episodes of abdominal pain, a variety of other abdominal complaints, electroencephalogram abnormalities, and response to anti-epileptic agents. We described the clinical profile of six patients with AE.**

Methods: **We conducted a retrospective survey of AE in children from the records of the hospital. The diagnosis of AE was dependent on recurrent abdominal symptoms, subtle central nervous system abnormalities, electroencephalogram abnormalities and response to anticonvulsant agents.**

Results: **The six patients were diagnosed with AE. The incidence of the disease was 0.07% in all admissions to the pediatric ward. Recurrent pain was common in all patients except two who had additional recurrent vomiting. In this series, the boy to girl ratio (1:2) was unequal.**

Conclusion: **High suspicion is required for the diagnosis of AE after exclusion of other possible causes.**

World J Pediatr 2014;10(1):80-82

Key words: **abdominal pain;**
children;
epilepsy

Brief report

Introduction

Abdominal epilepsy (AE) is an uncommon disorder in which abdominal pain along with a variety of other gastrointestinal complaints results from seizure activity.[1] It is characterized by unexplained, paroxysmal gastrointestinal complaints, subtle symptoms of a central nervous system disturbance, an abnormal electroencephalogram (EEG), and improvement with anticonvulsant drugs.[1,2] The common gastrointestinal symptoms include abdominal pain, nausea and vomiting, and the most frequent neurological abnormalities are lethargy and confusion. After exclusion of common etiologies, workup should proceed with an EEG. Management with anticonvulsant medications for resolution of symptoms confirms the diagnosis.[3,4] The exact etiologies of AE are not known.[1-4]

Methods

In the present retrospective survey of clinical data from the records of the hospital, 8432 patients were admitted to the Department of Pediatric Medicine during seven years (2003-2009). Among them, 6 patients had AE. They were diagnosed by recurrent abdominal symptoms, subtle central nervous system abnormalities, consistent abnormalities on EEG and response to anticonvulsant agents.

Results

In this present series of six cases, the incidence of AE was 0.07% of all cases admitted to the pediatric ward. Most of them were of older age group between 8-12 years (Table). Case 1 and 4 presented at a an early age. The mean age was 7.58 years. Among them, four patients were females and two males. Only one of them had a family history of epilepsy (case 1) and one had Down syndrome (case 6). All patients had visited Pediatric Emergency and Out-patients Department several times, but none of them visited Neurology Clinic. Duration from the onset of first symptom to confirmation of final diagnosis was short in most patients ranging from 6 to 9 months, except in case 3 with 7 years.

All had common symptoms of paroxysmal abdominal pain. Three patients (case 1, 3 and 5) had recurrent vomiting in addition to pain abdomen, who were diagnosed as recurrent abdominal pain due to functional gastro-intestinal abnormalities. They were treated with antiemetic, ante-reflux medication, antacids, antiamebics, antihelminthics or diazepam,

Author Affiliations: Department of Pediatrics, North Bengal Medical College & Hospital, Siliguri, India (Mondal R, Bag T, Mondal K, Saren A); Department of Pediatrics, Institute of Post Graduate Medical Education & Research, Kolkata, India (Sarkar S)

Corresponding Author: Rakesh Mondal, Balarampur, Mahestala, Kolkata-700141, India (Tel: +919674240973/033-64143736; Email: ivanrakesh2001@gmail.com)

doi: 10.1007/s12519-014-0457-1

Table. Summary of cases with abdominal epilepsy

Case	Age	Sex	Symptom duration	Clinical presentations	Physical examination	Investigations	EEG features	Medication and outcome
1	2.5 y	F	6 mon	Episodic abdominal pain and occasional vomiting	No physical abnormality, no neuro-deficits	Hemogram, serum electrolytes, urine, stool RE, X-ray, USG abdomen, UGI endoscopy, MRI brain, all normal	Paroxysms of sharp and slow waves suggestive of seizure disorder	Carbamazepine was started and continued for 2 y. Abdominal symptoms subsided. Symptom-free on 5 y follow-up.
2	9 y	M	9 mon	Localized epigastric pain for few minutes aggravation with a brief episode of unconsciousness and incoherent speech	Nothing contributory	Hemogram, urine and stool RE, chest X-ray, X-ray, USG, CT abdomen, UGI endoscopy, CT, MRI brain, all normal; Urine PBG, negative	Pattern consistent with of generalized cerebral dysrhythmia	Put on phenytoin sodium for 2 y. Symptom-free on 3 y follow-up. No recurrence of symptom.
3	12 y	F	7 y	Recurrent episodes of vomiting with abdominal pain (10-12 times per day) followed by headache and dizziness	No significant findings. No neuro-deficits	Hemogram, electrolytes, chest X-ray, USG abdomen, UGI endoscopy, CT, MRI of brain all normal. Urine PBG, negative	Focal temporal seizure with generalization, diffuse cortical slowing in other areas	Sodium valproate was started. She remains symptom-free on regular follow-up for two years. Valproate discontinued.
4	2 y	M	6 mon	Admitted in drowsy state and fixed tonic posture following an episode of pain abdomen. Similar attacks earlier	Drowsiness is transient and neurological examination in between-normal	Hemogram, electrolytes, chest X-ray, USG abdomen, UGI endoscopy and neuro imaging, all normal	High voltage generalized slowing	Phenytoin sodium was started. Abdominal pain and neurological symptoms subsided. Phenytoin was discontinued after 2 y.
5	12 y	F	6 mon	intermittent peri-umbilical pain and vomiting associated with drowsiness lasting for few minutes	Child was normal on general and systemic physical examination	A few small mesenteric nodes on USG abdomen. FNAC of the nodes revealed non-specific reactive hyperplasia. Other tests are normal	Awake inter-ictal mixed background rhythm with occasional sharp and slow wave pattern	She was put on sodium valproate. Free of symptoms on follow-up for last one year.
6	8 y	F	6 mon	Pain abdomen, nausea for 5-10 min followed by altered sensorium and fixed tonic posture 4 episodes previously, twice of admission	No significant findings except features of Down syndrome. No neuro-deficits	Hemogram, urine and stool RE, X-ray, USG abdomen, UG I endoscopy, all normal. MRI brain, no abnormality	Temporal lobe seizure with sharp and slow electrical discharge	Carbamazepine was started. She remained free of symptoms over next 3 mon.

F: female; M: male; RE: routine examination; USG: ultra sonography; UGI: upper gastro-intestinal; PBG: porphobilinogen; CT: computerized tomography; MRI: magnetic resonance imaging; EEG: electroencephalogram; FNAC: fine needle aspiration cytology.

which provided temporary relief. All of them had certain neurological symptoms presenting with unconsciousness lasting for few seconds to few minutes following episodes of pain abdomen. Fixed tonic posture was noticed in two cases (case 4 and 6). Case 2 had incoherent speech. Systemic examination was unremarkable in all, and no neuro-deficit was found.

They were investigated thoroughly for their gastro-intestinal symptoms to rule out any organic disease. Complete hemogram, routine examination of urine and stool, serum electrolytes, urea, creatinine were normal in all patients. X-ray of chest and abdomen were non-contributory. Mantoux test was negative in all. Sonography of the abdomen was unremarkable in all except in case 5, who was detected with mesenteric lymph nodes. However, fine needle aspiration cytology from these glands revealed a nonspecific reactive hyperplasia. Computerized tomography (CT) of the abdomen and upper gastrointestinal endoscopy were normal in all patients. Urinary porphobilinogen, done in two patients (case 2 and 3), whose symptoms simulated acute intermittent porphyria, were negative. Neuro- imaging in the form of CT brain and magnetic resonance imaging (MRI) of the head were non-contributory. All patients had demonstrable and distinctly consistent abnormalities on EEG suggestive of seizure disorders. Carbamazepine (case 1 and 6), valproate (case 3 and 5) and phenytoin (case 2 and 4) were used as anticonvulsants, and all of them showed good results. All patients were monitored at the Out-patients Department. Anticonvulsants were discontinued in most cases after 2 years of symptom-free follow-up. Abdominal or neurological symptoms did not recur in any of the patients.

Discussion

AE is an extremely rare entity in children. Approximately 36 patients including adults and children have been

reported in the literature. Gastro-intestinal complaints, commonly abdominal pain, were due to seizures.[1-4] The paucity of reports may be attributable to failure to recognize this ill-defined condition and inadequate documentation.

Clinical and epidemiological data from the Indian subcontinent especially in the pediatric subpopulation are few. Dutta et al[3] reported four cases of AE from India, including adolescents. Acute intermittent porphyria is a rare condition characterized by abdominal pain and a variety of nonspecific symptoms.[5] Its diagnosis is made by appropriate tests whenever required. In our study, porphyria was ruled out in two children. Syringomyelia is another condition that mimics AE. Patients usually have a gait disorder and muscle weakness in addition to recurrent abdominal pain.[6] In our patients, such abnormalities were not detected by MRI.

Gastro-intestinal disturbances in AE include recurrent abdominal pain, nausea, recurrent vomiting and diarrhoea, or their combinations.[1-3] Paroxysmal abdominal pain was seen in two of our patients and recurrent vomiting along with abdominal pain in another two patients. Patients with recurrent vomiting did not meet the criteria of cyclic vomiting. The frequency of vomiting in cyclic vomiting was three attacks lasting for one hour to 10 days in a period of 6 months. In each attack, vomiting occurred four times per hour for at least one hour without other attributable etiology as per consensus statement of North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition. The frequencies of vomiting in our patients were not as many as described.[7] Central nervous system (CNS) symptoms are usually subtle, confusion, fatigue, headache, dizziness, syncope, and lethargy. A family history of epilepsy may give a clue to the diagnosis of AE. Disorientation during an episode of pain followed by exhaustion and sleep may suggest AE in most of cases.[2,3] EEG as a simple, non-invasive method may be useful in differentiating AE from other diagnoses.[3,4] Patients with AE have temporal lobe seizures, generalized spike,wave discharges and frontal lobe discharge.[3,4] EEG features are not always consistent. In our patients, there were EEG abnormalities.

One of the accepted criteria for the diagnosis of AE is sustained response to anticonvulsants that include carbamazepine, phenytoin and sodium valproate.[1-4] Two of our patients received phenytoin, carbamazepine, sodium valproate. The anticonvulsants were tolerated well by all patients. In the patients, symptoms were controlled within one month after treatment.

In conclusion, in the diagnosis of AE subtle CNS symptoms in the presence of paroxysmal episodes of abdominal pain and recurrent vomiting should alert clinicians who are not neurologists. Specific question should always be asked regarding the neurological symptoms. Treatment with an anticonvulsant is usually effective. Further research is necessary to generate clinical and epidemiological data on risk factors, etiologies and genetic predisposition for proper management.

Funding: None.
Ethical approval: Not needed.
Competing interest: None.
Contributors: MR, BT were involved in diagnosis and management of the patients. MR and SS searched the literature. MR, MK, SS and SA drafted the manuscript. MR and SS did the critical review. All the authors approved the final version. MR and BT are guarantors.

References

1 Zinkin NT, Peppercorn MA. Abdominal epilepsy. Best Pract Res Clin Gastroenterol 2005;19:263-274.
2 Franzon RC, Lopes CF, Schmutzler KM, Morais MI, Guerreiro MM. Recurrent abdominal pain: when should an epileptic seizure be suspected? Arq Neuropsiquiatr 2002;60:628-630.
3 Dutta SR, Hazarika I, Chakravarty BP. Abdominal epilepsy, an uncommon cause of recurrent abdominal pain: a brief report. Gut 2007;56:439-441.
4 Ashir GM, Alhaji MA, Gofama MM, Ibrahim AB, Azuka NC. Abdominal epilepsy in a Nigerian child: A case report. S Afr J Child Health 2009;3:95-97.
5 Kuo HC, Huang CC, Chu CC, Lee MJ, Chuang WL, Wu CL, et al. Neurological complications of acute intermittent porphyria. Eur Neurol 2011;66:247-252.
6 Drexler DL, Grill BB, Ashwal S. Spinal cord tumor-associated syrinx mimicking abdominal epilepsy: a rare cause of childhood abdominal pain. J Pediatr Gastroenterol Nutr 1989;9:524-527.
7 Li BU, Lefevre F, Chelimsky GG, Boles RG, Nelson SP, Lewis DW, et al. North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition consensus statement on the diagnosis and management of cyclic vomiting syndrome. J Pediatr Gastroenterol Nutr 2008;47:379-393.

Received September 11, 2012
Accepted after revision May 10, 2013