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ICTUS EMETICUS: CASE REPORTS AND LITERATURE REVIEW

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ABSTRACT

The diagnosis of abdominal epilepsy came into vogue in the 1950s and 1960s. Vomiting as a manifestation of seizure has been given different names including ictus emeticus. We report three cases of this interesting albeit uncommon condition. It is important for physicians to familiarize themselves with this symptomatology so as not to overlook this unique presentation of epileptic seizures.

Vomiting as a manifestation of partial seizures has been long recognized, described by various names such as ictus emeticus, cyclic vomiting, and abdominal epilepsy.¹ Abdominal epilepsy, as described in the literature, is manifested as various gastrointestinal symptoms including epigastric distress, peculiar 'rising sensation', nausea, retching, vomiting, salivation and borborygmi.²⁻⁵ The incidence of ictus emeticus has been reported from 1.9% to 2.7% in large series of patients with temporal lobe epilepsy.^{4,6} The localizing value of vomiting is unclear. No sex or race predisposition has been reported. We present three patients whose clinical picture is suggestive of ictus emeticus.

CASE REPORTS

Case 1. A 12-year-old boy with Fragile-X syndrome and attention deficit hyperactivity disorder (ADHD) developed seizures that would begin with the patient complaining 'I feel sick, I am sick'. He appeared pale, and gagged as if he was going to vomit. These spells would last for a few minutes and he would subsequently be incoherent and disoriented. He also experienced other spells that were described as eyes rolling back, becoming incoherent and having urinary incontinence. At the time of first evaluation, he was on divalproex sodium 250 mg every morning and 500 mg every evening, in addition to methylphenidate 10 mg three times daily. He was having multiple seizures in a month, sometimes up to 8 or 9 in a day. EEG showed diffuse slowing with no epileptiform discharges. He was

switched to an extended-release form of carbamazepine at the dosage of 400 mg twice a day, and his seizure frequency decreased to under 2 per month.

Case 2. An 18-year-old man was seen with a history of seizures since the age of 6. His initial seizures comprised of episodes of loss of contact from surroundings, staring into space, and vomiting. He had an abnormal EEG at age 6 and was started on phenytoin with good control of his spells. He remained on phenytoin for about 2 years and thereafter discontinued. At the age of 18, he began to experience vomiting five to thirty minutes after eating. Initially infrequent, these vomiting episodes became a daily occurrence. After the vomiting he appeared sleepy and at times would stare into space. A complete gastrointestinal work up to determine the etiology of these vomiting spells was non-revealing. He was put on promethazine and pantoprazole, which failed to stop the vomiting. When he presented for neurological evaluation, he was having almost daily episodes of vomiting. Neurological examination was normal. An EEG showed bilateral frontal spikes, with sharp and slow-wave complexes. He was started on phenytoin, which improved his spells and he stopped having daily vomiting. Phenytoin led to toxicity and was switched to divalproex sodium. His vomiting recurred and he was put back on phenytoin on a lower dose, on which the spells appeared to be controlled. In a subsequent follow-up visit, he reported recurrence of the vomiting spells. Phenytoin level at the time was 3.8 µg/ml. Increase in phenytoin dosage resulted in remission of vomiting spells.

Case 3. A 50-year-old woman, known to have diabetes, congestive cardiac failure and pulmonary hypertension, presented with a 2-month history of unexplained episodic vomiting, diarrhea and abdominal pain, associated with loss of consciousness or decreased responsiveness. Events started with nausea and vomiting followed by a brief period of loss of consciousness or awareness during which the patient did not respond or talk. The frequency of these episodes ranged from several per day to an episode-free period lasting two days. Neurological examination was normal. The patient had a complete gastrointestinal workup to determine the etiology of these symptoms but the investigations proved non-revealing apart from a mild gastritis that was insufficient to explain the patient's symptoms. A brain CT scan was normal, although an EEG showed left temporal spikes. She was started on phenytoin 100 mg twice daily and responded well: within three days of starting treatment all symptoms including nausea, vomiting, and altered mentation showed improvement. On a recent follow-up visit, her symptoms remained subsided, and no signs of phenytoin toxicity were apparent.

DISCUSSION

Recurrent vomiting that occurs in a paroxysmal fashion and which cannot be assigned to another pathology has been implicated as a manifestation of epileptic seizures.⁶ It is associated with more than one type of epilepsy, with the highest frequency seen in simple partial seizures.⁷ All three of our patients had symptoms suggestive of ictus emeticus. Although there is a high preponderance of ictus emeticus in benign childhood epilepsy, one of our patients was an adult female, which suggests that abdominal epilepsy is not limited to the pediatric age group.

A variety of symptoms have been reported as manifestations of ictal vomiting. Commonly reported manifestations of ictus emeticus include paroxysmal abdominal pain, retching, belching, cough, nausea, vomiting, and salivation.^{1,4,8} Approximate duration is from 10 to 38 seconds.³ Chen et al⁴ further classified vomiting characteristics into retching, retching with ejection of gastric contents, and projectile vomiting without retching. These symptoms may or may not be associated with altered awareness and disorientation. These seizures may secondarily generalize.^{6,9} Amnesia for the event has been frequently reported.⁶ Kaplan et al¹⁰ have reported a case of ictal spitting (ictus expectoratus) in a patient with right temporal lobe ganglioglioma who responded to resection. They suggested involvement of the non-dominant temporal lobe as the main area responsible for producing vomiting and spitting during an ictal episode. A similar report pertaining to ictal spitting is also presented by Kellinghaus

et al.¹¹ Mitchell et al¹² have reported a case of astrocytoma in the right hemisphere associated with seizures comprised of olfactory hallucinations and vomiting. Since vomiting is also a feature of raised intracranial pressure and CNS infections, this condition can be misdiagnosed mainly as encephalitis or other serious insults.¹³

Our patients responded well to the traditional anti-epileptic medications phenytoin and carbamazepine. Case 2 experienced recurrence of vomiting with sub-therapeutic levels of phenytoin, and an increase in phenytoin dose resulted in remission. There is no single preferred anti-epileptic medication for this kind of seizures and most of the older anti-epileptic medications have been reported in the management of these seizures. A few authors, however, have preferred phenytoin,² and certainly phenytoin proved to be most effective in our patients. Interestingly, our patients did not show a good response to divalproex sodium. One drawback to phenytoin was the appearance of toxicity early on during treatment, which may limit its use. The traditional medications used for the treatment of partial seizures have been reported to be effective but have limitations due to side effects. We found no reports of treating this condition with the newer antiepileptic medications that have better safety profiles.

The exact mechanism of ictal vomiting remains unknown.^{4,14} There is evidence of an insular trigger.⁹ The possible mechanism involves the spread of abnormal activity through descending insular or limbic circuits.⁷ Localization has been implicated at the lateral and superior region of the temporal lobe and the insular cortex, which contains a neural map of the viscera.⁴ Some authors, suggesting hemispheric asymmetry of gastrointestinal control, localize ictal vomiting to the non-dominant hemisphere.^{1,5,10,15} However, other authors have argued against it.^{4,16} Most authorities agree on right temporal involvement, yet there has been strong evidence presented of left temporal lobe involvement.¹⁷ Schindler et al¹⁸ reported a case of ictal vomiting in left temporal lobe epilepsy. Fiol et al⁹ demonstrated a possible role for insular cortex as a trigger of vomiting in their patient with ictus emeticus. Guerrini et al¹⁵ have reported patients whose seizures started in the occipital lobe and spread to the temporal lobe and the patients then experienced retching or vomiting. Some authors have reported that ictal vomiting can be induced by intermittent photic stimulation (IPS).^{15,19}

Paroxysmal vomiting in childhood has been attributed either to migraine or seizures. An epileptic syndrome (Panayiotopoulos Syndrome) has been recognized in which vomiting is part of the seizure phenomenon.^{13,20,21} In this syndrome, also known as early-onset childhood epilepsy

with occipital spikes (Panayiotopoulos-type), seizures are comprised of autonomic and behavioral disturbances, vomiting, deviation of the eyes, and often impairment of consciousness that can progress to convulsions. In this syndrome, the most common presentation was that of ictus emeticus at onset followed by deviation of the eyes or staring, loss of contact, and floppiness.²² Kivity et al²⁰ have reported vomiting in less than 50% of their patients with this syndrome. Recent studies have suggested a link between Rolandic epilepsy and Panayiotopoulos syndrome, the two most common phenotypes of benign childhood seizure susceptibility syndromes.²²

EEG is critical for the diagnosis according to Shuper et al.⁷ Our patients also showed EEG abnormalities such as bilateral frontal sharps, and temporal spikes which led us to establishing the diagnosis of ictus emeticus.

Vomiting as a manifestation of simple partial or complex partial seizures is not too uncommon. It is probably under-recognized. Physicians, especially neurologists, should be more cautious while taking a history of seizures, as patients may not volunteer vomiting as a symptom of their seizures. Though evidence favors ictal vomiting arising from the non-dominant hemisphere, this is not the rule. While performing EEG in patients suspected to have ictal vomiting, intermittent photic stimulation should be done. There is no single preferred treatment for this manifestation of epilepsy; however, most of the older medications used in the treatment of partial epilepsy are effective.

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