

Case Report

Abdominal Epilepsy: An Uncommon of Non-Convulsive Status Epilepticus

Somsak Tiamkao MD*,
Thongchai Pratipanawat MD**, Suthipun Jitpimolmard MD*

* Division of Neurology, Department of Medicine, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

** Division of Endocrinology, Department of Medicine, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

Objective: To recognize abdominal epilepsy in adults.

Material and Method: Case report.

Results: Case 1: A 21-year-old woman with DM type I presented with a 2-month history involving four episodes of severe abdominal pain and vomiting, each of which lasted four to five days. She had a recurrence every two weeks. The EEG revealed 'spike and wave' and she was started an intravenous phenytoin that resolved the symptoms. Case 2: A 20-year-old woman with DM type I was admitted with a 2-month history of recurring severe left upper quadrant pain associated with occasional nausea but no vomiting. She experienced two more episodes of generalized tonic-clonic seizures and was treated with 300 mg phenytoin given orally. The abdominal pains gradually subsided and she was symptom-free within two days. An EEG showed frequent sharp waves. She was treated with 10 mg intravenous diazepam and her symptoms and sharp waves disappeared within two minutes. Case 3: A 46-year-old man with DM type I was admitted with a four-month history of recurring severe epigastric pain and vomiting. His physical examination, laboratory tests, and extensive investigation for a primary GI disorder revealed nothing unusual. The EEG revealed spike and wave and he was treated with intravenous AED (phenytoin) loading after which the symptoms disappeared.

Conclusion: Physicians should consider abdominal epilepsy in diabetics with recurrent, intractable abdominal pain in whom extensive investigations for primary gastrointestinal (GI) disorders are unremarkable.

Keywords: Abdominal epilepsy, Non-convulsive status epilepticus, Abdominal pain

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Abdominal epilepsy is an uncommon cause of gastrointestinal complaints in adults. Such were first reported in 1868 by Trousseau, in a boy with paroxysmal attacks of abdominal pain, vomiting, giddiness, pallor, who later developed grand mal attacks⁽¹⁾. Most reported cases are in children and adolescents. Abdominal epilepsy is usually treated with an antiepileptic drug (AED). Clinical manifestations include abdominal pain, most commonly sharp or colicky (86%), nausea and/or vomiting (28%), diarrhea (5%), and bloating (3%)⁽²⁾. The site of abdominal pain is most commonly periumbilical or in the upper abdomen. Many patients have associated disturbances of consciousness and an abnormal electroencephalogram (EEG). Although

the abdominal symptoms may be similar to those of irritable bowel syndrome, they may be distinguished from the latter by the occurrence of altered consciousness during the attacks, tiredness after an attack and/or by an EEG positive for epileptic waveform. This unusual cause for abdominal pain should be considered in patients with abdominal symptoms who do not respond to the usual treatment.

The authors' objective was to present three cases reports of diabetic patients who suffered from abdominal pain for several months before being diagnosed with abdominal epilepsy.

Case Report

Case 1

A 21-year-old woman with DM type I presented with a two-month history characterized by four episodes of severe abdominal pain and vomiting, each of which persisted for four to five days. During

Correspondence to:

Tiamkao S, Division of Neurology, Department of Medicine, Faculty of Medicine, Khon Kaen University, Khon Kaen 40002, Thailand.

Phone: 043-363-664, Fax: 043-374-542

E-mail: somtia@kku.ac.th

each attack, she could not eat and needed to have stomach contents (1,500 cc/day) removed with a nasogastric tube.

Her physical examination, laboratory tests and extensive evaluation for a primary GI disorder were unremarkable (including stool exams for ova and parasites, abdominal ultrasound, routine blood exams, such as liver, pancreas, renal function, upper and long GI study, and gastro scope).

She had a recurrence every two weeks. An EEG during an attack revealed a spike and wave at both temporal lobes (Fig. 1). She was then started on intravenous phenytoin, which abolished the symptoms, then the her brain MRI was normal.

Case 2

A 20-year-old woman with DM type I was admitted with a two-month history of recurring severe left upper quadrant pain associated with occasional nausea but no vomiting. The abdominal pains were associated with chest discomfort and dizziness. She experienced two episodes of generalized tonic-clonic seizures (GTCs) and was treated with phenytoin (300 mg) orally. The abdominal pain gradually subsided and she was symptom-free within two days. Notwithstanding, three weeks later, she developed an episode of severe abdominal pain and an EEG showed diffuse frequent sharp waves (Fig. 2). She was treated with intravenous diazepam (10 mg) and her symptoms and sharp waves disappeared within two minutes. A subsequent brain MRI was normal.

Case 3

A 46-year-old man with DM type I was admitted with a four-month history of recurring severe epigastric pain and vomiting. His physical examination, laboratory tests and extensive investigation for a primary GI disorder revealed normal findings. He had recurrent episodes monthly. An EEG revealed spike and wave, so he was treated with intravenous phenytoin loading and the symptoms disappeared. His brain MRI was normal.

Discussion

Non-convulsive status epilepticus (NCSE) refers to prolonged seizure activity in the absence of major motor signs⁽³⁾. The four major types of NCSE are absence status epilepticus, simple partial status epilepticus (SPSE) without motor features, complex partial status epilepticus (CPSE) and status epilepticus in coma including subtle status epilepticus.



Fig. 1 An EEG revealed a spike and wave at both temporal lobes

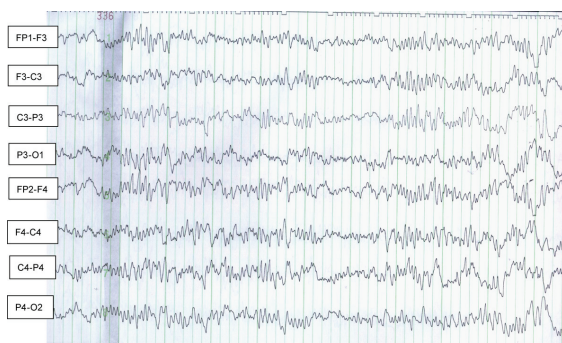


Fig. 2 An EEG showed diffuse frequent sharp waves

NCSE is described as a change in behavior and/or mental processes from baseline associated with continuous epileptic-form discharges on the EEG. Nonetheless, there is no universally-accepted definition of NCSE. Some suggested definitions include different components such as clinical changes that usually incorporate impaired consciousness, ictal EEG abnormalities, and response to treatment. Therefore, saying, the definition must not exclusively rely on EEG changes because no single pattern can be regarded as pathognomonic. A positive electro clinical response to acute AED treatment may be helpful in the diagnostic process, but 'no response' does not exclude its diagnosis.

Abdominal sensations commonly herald the onset of a motor seizure. This so-called abdominal aura, which may include sensations such as pain, nausea, gassiness and hunger, is the most common aura in epilepsy, especially temporal lobe epilepsy. However, gastrointestinal signs and symptoms may be the primary or only manifestation of seizures hence, abdominal epilepsy.

The diagnostic criteria for abdominal epilepsy include (1) paroxysmal gastrointestinal complaints unexplained by complete evaluation (including laboratory tests, radiographic imaging, and endoscopy), (2) symptoms of a CNS disturbance, (3) an abnormal EEG with findings specific for a seizure disorder and (4) a sustained abolition of symptoms with AED⁽²⁾. The diagnosis of abdominal epilepsy begins with eliciting a history typical for the syndrome, namely paroxysmal and brief episodes of abdominal pain or other gastrointestinal symptoms. The presence of associated neurological symptoms including convulsions, impaired consciousness, and other sensory phenomenon, is an important clue and should be elicited when considering the diagnosis, albeit neurological symptoms may not be present with every episode.

An appropriate evaluation including physical and neurological examination, laboratory studies, endoscopy, and abdominal imaging, usually with computed tomography and ultrasound, should be performed to exclude other more common etiologies. Then, when none is found, or the history is still suggestive, an EEG should be performed to look for abnormalities specific to epilepsy. These so-called interictal, epileptiform discharges (IEDs) comprise paroxysmal spikes and/or sharp waves. There are no studies on the sensitivity of EEG for this condition and but a few from case series showing the range of positive EEGs (29-55%). An EEG recorded during an event can be normal, even if there is no associated convulsion or impairment of consciousness⁽⁴⁾.

How seizures induce abdominal sensations has been studied but the etiology is still unclear. Stimulation of the temporal lobe structures, including sensations, hippocampus, and insular cortex, has been shown to induce abdominal sensations, including nausea, hunger and 'funny feelings', in humans⁽²⁾. Some researchers have postulated that abdominal symptoms could result from true visceral stimuli which, through connections to the brain, could induce seizures⁽⁵⁾. The first case usually developed symptoms of dyspepsia, nausea/ vomiting or diarrhea and then followed with abdominal epilepsy. Most cases of abdominal epilepsy are of temporal lobe origin⁽⁵⁾ although several cases have also been traced to parietal lobe lesions⁽⁶⁾. An EEG of the first case found spike and wave at both temporal lobes (Fig. 1).

Treatment usually starts with an AED, typically phenytoin. Importantly, no controlled trials have been published regarding treatment of abdominal

epilepsy. Moreover, while helpful for confirming a diagnosis of abdominal epilepsy, a response to therapy alone is not diagnostic. Therefore, AED therapy may improve many non-epileptic causes of abdominal pain, either by its sedating effect or as a placebo.

Hyperglycemia may present with seizures, visual hallucination, abnormal movement (chorea, hemiballism) and coma in severe cases of hyperglycemia^(7,8). Hyperglycemia induced seizure are usually focal seizures⁽⁸⁾ and epilepsia partialis continua (EPC)⁽⁹⁾. Seizures are more common in non-ketotic compared to ketotic hyperglycemia. Keto-acidosis decrease neuronal excitability by increasing level of GABA⁽¹⁰⁾. Tiamkao S⁽¹¹⁾ reported 21 patients with non-ketotic hyperglycemic induced seizures (NKHS), EPC were found in 80%. NKHS usually controlled with insulin therapy, lowering the plasma glucose less than 200 mg/dL usually controls NKHS and no AED is need^(11,12). All of the three cases in the present report had underlying DM type I, treated with insulin therapy. All of admissions, their plasma glucose level were higher than 500 mg/dL and no metabolic acidosis. All of patients need to control plasma glucose and IV AED for control abdominal epilepsy. All three cases had a history of high plasma glucose, which precipitated abdominal epilepsy.

All three of our patients had recurrent episodes of severe abdominal pain, each episode lasting for 4-5 days. All GI investigations and blood tests were unremarkable and an EEG during an attack showed abnormal findings specific for a seizure disorder. Sustained abolition of symptoms was achieved with AED. Since all of the clinical manifestations were compatible with NCSE⁽³⁾, the presented patients were diagnosed with abdominal epilepsy.

Conclusion

Physicians should consider abdominal epilepsy in diabetic patients with recurrent, intractable abdominal pain and in whom extensive investigations of primary GI disorder were unremarkable.

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Potential conflicts of interest

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โรคลมชักชนิดปวดท้อง: ภาวะชักต่อเนื่องชนิดไม่กระตุ้นที่พบไม่บ่อย

สมศักดิ์ เทียมเก่า, ธงชัย ประภาณวัตร, สุทธิพันธ์ จิตพิมลมาศ

วัตถุประสงค์: ทราบถึงโรคลมชักชนิดปวดท้องในผู้ใหญ่

วัสดุและวิธีการ: รายงานผู้ป่วย

ผลการศึกษา: รายที่ 1 ผู้หญิงอายุ 21 ปี ป่วยเป็นโรคเบาหวานชนิดที่ 1 มีประวัติปวดท้องอย่างรุนแรงร่วมกับอาเจียน 4 ครั้งในระยะเวลา 2 เดือน อาการแต่ละครั้งนาน 4-5 วัน ผู้ป่วยมีอาการเป็นไข้ทุก 2 สัปดาห์ ผลการตรวจคลื่นไฟฟ้าสมองพบ spike และ wave ได้รับการรักษาด้วยยาฟีนัยโตอินฉีดเข้าทางหลอดเลือดดำและอาการหายไป รายที่ 2 ผู้ป่วยหญิงอายุ 20 ปี ป่วยเป็นโรคเบาหวานชนิดที่ 1 รับไว้รักษาในโรงพยาบาลด้วยประวัติปวดบริเวณช่องท้องด้านซ้ายบนเป็นซ้ำ บางครั้งมีอาการคลื่นไส้แต่ไม่อาเจียน ผู้ป่วยเคยชักแบบเกร็งกระตุกทั้งตัว 2 ครั้ง และรักษาด้วยยาฟีนัยโตอิน 300 มก./วัน ชนิดรับประทาน อาการปวดท้องค่อย ๆ ลดลง และหายภายใน 2 วัน ผลการตรวจคลื่นไฟฟ้าสมองพบคลื่นสมองลักษณะ sharp wave บ่อย ๆ ผู้ป่วยรักษาด้วยยาไดอะซีแพมชนิดฉีดเข้าทางหลอดเลือดดำ อาการ และคลื่นสมองลักษณะ sharp wave หายไปภายใน 2 นาที รายที่ 3 ผู้ป่วยอายุ 46 ปี ป่วยเป็นโรคเบาหวานชนิดที่ 1 รับไว้รักษาในโรงพยาบาลด้วยประวัติ ปวดบริเวณลิ้นปี่อย่างแรง และอาเจียนซ้ำ ๆ ตรวจร่างกาย ผลการตรวจทางห้องปฏิบัติการและการตรวจเฉพาะโรคทางระบบทางเดินอาหาร ไม่พบความผิดปกติ ผลการตรวจคลื่นไฟฟ้าสมองพบ spike และ wave ผู้ป่วยได้รับการรักษาด้วยยากันชักฟีนัยโตอินฉีดเข้าทางหลอดเลือดดำแบบโคม หลังจากนั้นอาการปวดบริเวณลิ้นปี่หาย

สรุป: แพทย์ควรคิดถึงโรคลมชักชนิดปวดท้องในผู้ป่วยโรคเบาหวานมีอาการปวดท้องซ้ำ ๆ ไม่ตอบสนองต่อการรักษา และได้รับการตรวจทางห้องปฏิบัติการโรคทางเดินอาหารไม่พบความผิดปกติ