

An Unusual Cause of Recurrent Paroxysmal Abdominal Pain in Adults (Abdominal Epilepsy)

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Paroxysmal, recurrent abdominal pain in adults is uncommonly caused by abdominal epilepsy, which is one spectrum of temporal lobe epilepsy (TLE). It may be distinguished from the irritable bowel syndrome or migraine by the presence of central nervous system complaints including transient loss of consciousness, by response to antiepileptic medication and by epileptiform discharges in EEG. In this report, we review the variety of gastrointestinal symptoms and central nervous system disturbances, specific encephalographic abnormalities and abnormal brain MRI findings of abdominal epilepsy. A striking response to anticonvulsant medication was seen in all adult patients.

Key word: abdominal epilepsy, recurrent paroxysmal abdominal pain, TLE

INTRODUCTION

A common, and often compounding, diagnostic challenge is the unfortunate person with recurrent abdominal pain. When a searching history, thorough physical examination, laboratory evaluation, and radiological studies are not available, such patients are often labelled neurotic, or advised to undergo exploratory surgery. It is in such a setting that the "abdominal epilepsy" is occasionally raised. Experimental studies in human have shown that stimulation of certain areas such as the amygdala, hippocampus, parahippocampus, temporal and extratemporal gyrus can evoke gastric and epigastric sensations¹. Up to 20% of patients with convulsive disorders, particularly those with temporal lobe epilepsy (TLE) will experience a viscer-

al aura such as epigastric distress, nausea, vomiting, borborygmi and belching^{2,3}. On occasion, however, these visceral symptoms may be the only sign of paroxysmal disturbance of cerebral electrical activity and, as such, have been labelled as convulsive equivalent states^{1,4}. An example of this is abdominal epilepsy. Abdominal epilepsy is an uncommon cause of recurrent abdominal and/or other epigastric pain and central nervous system symptom. Typically, the pain is abrupt in onset, localized to the periumbilical or upper abdomen, and generally brief, uncommonly lasting more than a few minutes⁵. It is paroxysmal in nature and not chronic. It may be accompanied by anorexia, vomiting, or diarrhea and is frequently associated with a change in consciousness, such as disorientation or confusion, but is rarely associated with a brief

loss of consciousness. For the most part it is a disorder of children with very rare occurrence in adult patients^{6,7}. Our experience over the previous two years indicates that the disorder may not be as rare as is suggested by the paucity of literature on the subject. Moreover, the variability of the clinical presentation indicates a spectrum to both the gastrointestinal(GI) and central nervous system(CNS) manifestations of TLE in adults. This study reviews that spectrum and the clinical course in five patients.

PATIENTS AND METHOD

The charts of patients referred for GI evaluation between 1991 and 1993 were reviewed. Five patients fit the diagnosis of abdominal epilepsy. The initial criteria for the diagnosis was paroxysmal GI complaints unexplained by a complete evaluation

consisting of a thorough battery of laboratory tests, roentgenographic studies, ultrasound and/or abdominal CT scan, and in some cases, endoscopic evaluations. Other criteria included ; 1) symptoms of a CNS disturbance not explained by a structural abnormality on CT scan or MRI of the brain, 2) an abnormal electroencephalogram(EEG) with findings specific for a seizure disorder, and 3) a convincing sustained abolition of symptoms on anticonvulsant medication⁸.

RESULTS

All patients were Korean ranging in age from 16 to 50 yr, with an average of 38.8 yrs. The duration of symptoms prior to diagnosis varied from 8 months to 12 yrs, with an average length of 2.3yrs (see Table 1)

Table 1. Abdominal Epilepsy Patient Profile

Patient	Sex	Age(yr)	GI symptoms	CNS symptoms	Treatment
An, KJ	F	50	Epigastric distress, Indigestion, Bloating	Confusion, Restlessness, Neck deviation to left	Tegretol(600mg per day)
OK, BS	F	46	Nausea, Vomiting, Lower abdominal pain	Vertigo, Headache, Syncope, Sudden blindness	Tegretol(600mg per day)
Pak, YK	F	16	Paroxysmal periumbilical pain	Syncope, Headche	Phenytoin(300mg per day)
Lee, KH	M	50	Watery diarrhea, Urge to defecate, Lower abdominal pain	Syncope, Chest tightness, Febrile sensation	Phenytoin(30mg per day)
Choi, CB	F	32	Nausea, borborygmi, LtLUQ pain	Stiffness of face, Throbbing right temporal headache, Chest palpitation, Syncope	Tegretol(600mg per day)

Illustrative Cases

Case 3(Pak,YK) : A 16-yr-old girl presented with a 3-yr-history of recurring severe periumbilical pain associated with occasional nausea but no vomiting. The pains were associated with an overwhelming sense of fatigue such that she would

often lie down and fall asleep. At times, in association with the pain and fatigue, she had a severe throbbing headache in the bilateral temporal portion followed by syncope. Physical examination, laboratory tests and a through search for a primary GI disturbance were unremarkable. A sleep EEG showed bursts of generalized sharp waves

over the right fronto-temporal area. A brain MRI was unremarkable. She was started on phenytoin and for 15 months has been symptom free.

Case 4 (Lee, KH): A 50-yr-old man was examined for a 12-year history of frequent episodes of lower abdominal pain, watery diarrhea and severe sensation of tiredness in association with transient loss of consciousness for 1 to 2 minutes. Temporary relief was obtained by stooling. Precipitating causes for this discomfort and transient loss of consciousness were cold or spoiled food, especially at night. During the previous 2 yrs the frequency, duration, and intensity of the abdominal pain increased. He had two witnessed episodes of syncope, lasting 2 to 3 minutes each. Physical examination, laboratory tests, and evaluation for a primary GI disorder were unremarkable. A sleep EEG revealed frequent paroxysmal unsustained bursts of sharp waves most prominent over the bilateral temporal region. A brain MRI was normal. He was started on phenytoin with abolition of the episodes of diarrhea, paroxysmal lower abdominal pain and mental changes in spite of cold food intake at night. He has been controlled over a 1-yr period on phenytoin, 300mg/day.

Case 5 (Choi, CB): A 32-yr-old woman was referred for evaluation of 8 months of nausea, borborygmi, severe deep aching abdominal pain in the left upper quadrant to the epigastrium and a throbbing right temporal headache. Recently the frequency had increased up to 3/day with syncopal attack. Neurological and physical examination, laboratory tests, extensive GI workup (abdominal MRI, upper GI series, gastroscopy) and IVP study were unremarkable. An EEG was abnormal because of intermittent spikes over the left temporal (T5) areas (Fig. 1).

On tegretol she had complete relief of left upper quadrant pain, headache and brief loss of con-

sciousness. She has been controlled over the past 12-month with a dose of 600mg/day.

Gastrointestinal Symptoms

One patient experienced frequent episodes of paroxysmal periumbilical pain not associated with vomiting, or change in bowel function. Two patients reported prolonged lower abdominal pain, nausea, vomiting and/or watery diarrhea, and in one patient left upper quadrant pain associated with nausea and borborygmi. The remaining patient complained of severe nausea lasting several hours, indigestion associated with generalized bloating as though distended with gas.

Central Nervous System Symptoms

All patients experienced CNS symptoms occurring only in association with the GI complaints. Two patients reported several throbbing or dull aching headaches and/or vertigo. The headaches usually were pounding and unilateral and often lasted beyond the duration of the abdominal disturbance. One patient became confused with the onset of their abdominal symptoms and secondary generalized tonic seizure. Feelings of exhaustion and a need for prolonged sleep were experienced by one patient. Four patients had syncopal episodes and one of them had transient complete blindness, chest discomfort and febrile sensation during the paroxysm of gastrointestinal upset.

EEG Abnormalities

Each of the five patients had a specific EEG abnormality consisting of bursts of sharp waves and/or spikes over one or both temporal and frontal regions. Sleep studies were performed in four patients in order to optimize the conditions for de-

etecting any abnormality present.

MRI Findings

Atrophic hippocampal head and body with increased temporal horn or increased high signal on T2 weighted image was detected in three patients. Imaging was normal in the remaining two patients

Response to Medication

Anticonvulsant medication has resulted in prompt and the complete abolition of both GI and CNS disturbances in each of five patients in follow-up periods ranging from 8 months to 2 yrs. Two patients responded to phenytoin. Three patients are symptom-free on tegretol. Cessation of medication in two patients has led to recurrent abdominal pain and CNS symptoms and, therefore, patients have been educated to continue maintenance anticonvulsant therapy.

DISCUSSION

Abdominal epilepsy has been associated with a variety of abdominal complaints, including paroxysmal abdominal pain, nausea, vomiting, and diarrhea in children⁹, the described symptoms and signs include borborygmi, belching, vomiting, or flatus^{1,10-12} and defecation or the desire to defecate^{12,13}.

The few previous reports of temporal lobe epilepsy in adults have described periumbilical abdominal pain as the sole GI manifestation of the disorder⁷. The symptoms described in this report suggest that there is a spectrum of GI manifestations of TLE in adults which extends beyond periumbilical pain, and includes left upper quadrant and lower abdominal pain. They also had nausea, indigestion, bloating, borborygmi, flatus and watery diarrhea.

A variable diversity of CNS symptoms was noted

in these patients who presented with febrile sensation, chest tightness, throbbing or dull aching headache, vertigo, syncope, and transient loss of consciousness and blindness. This spectrum of CNS symptoms contrasts somewhat with those reported in children, who usually have only an impairment of consciousness with a desire for sleep⁵. But in Peppercorn's patients symptoms suggestive of CNS disturbance, such as, alteration of awareness, headache, syncope and transient blindness were also reported⁶. The relationship of abdominal migraine to temporal lobe epilepsy is not clear. Prichard believes that the two can be different on the basis of those with a migraine equivalent having a positive family history for migraine, a longer attack of pain, frequent associated headaches, and normal EEG¹⁴.

The type of change in electroencephalogram(EEG) was intermittent bursts of sharp waves or focal spikes, or both, often emanating from the temporal or frontal lobes. This finding is very different from previous cases, in a large percentage of those, a generalized dysrhythmia of slow activity was seen¹⁵. But similar epileptic discharges were reported in Peppercorn's patients⁵. Nearly fifty percent sensitivity(predominantly mesial temporal abnormalities) was reported with MRI study on TLE¹⁶. In our cases, three cases were abnormal on MRI(coronary view) among five patients. So this study is very meaningful for a workup of abdominal epilepsy.

The cause of abdominal epilepsy is uncertain and like other epilepsies, there may be numerous. It is also unclear whether the initial disturbance in abdominal epilepsy arises in the brain. There are direct sensory pathways from the bowel via the vagus nerve to the solitary nucleus of the medulla which is heavily connected to the amygdala¹⁷. These can be activated during intestinal contractions, which may be triggered by cold or spoiled food as in case 2(Lee, KH).

Having been diagnosed in only 5 patients referred for GI evaluation over a 2-yr period, the internist and neurologist should be alerted to the possibility of TLE in adult patients with unexplained paroxysmal abdominal pain associated with symptoms suggestive of a CNS disturbance. In such patients, a sleep EEG should be obtained and, if abnormal, a therapeutic trial of anticonvulsant medication should be considered.

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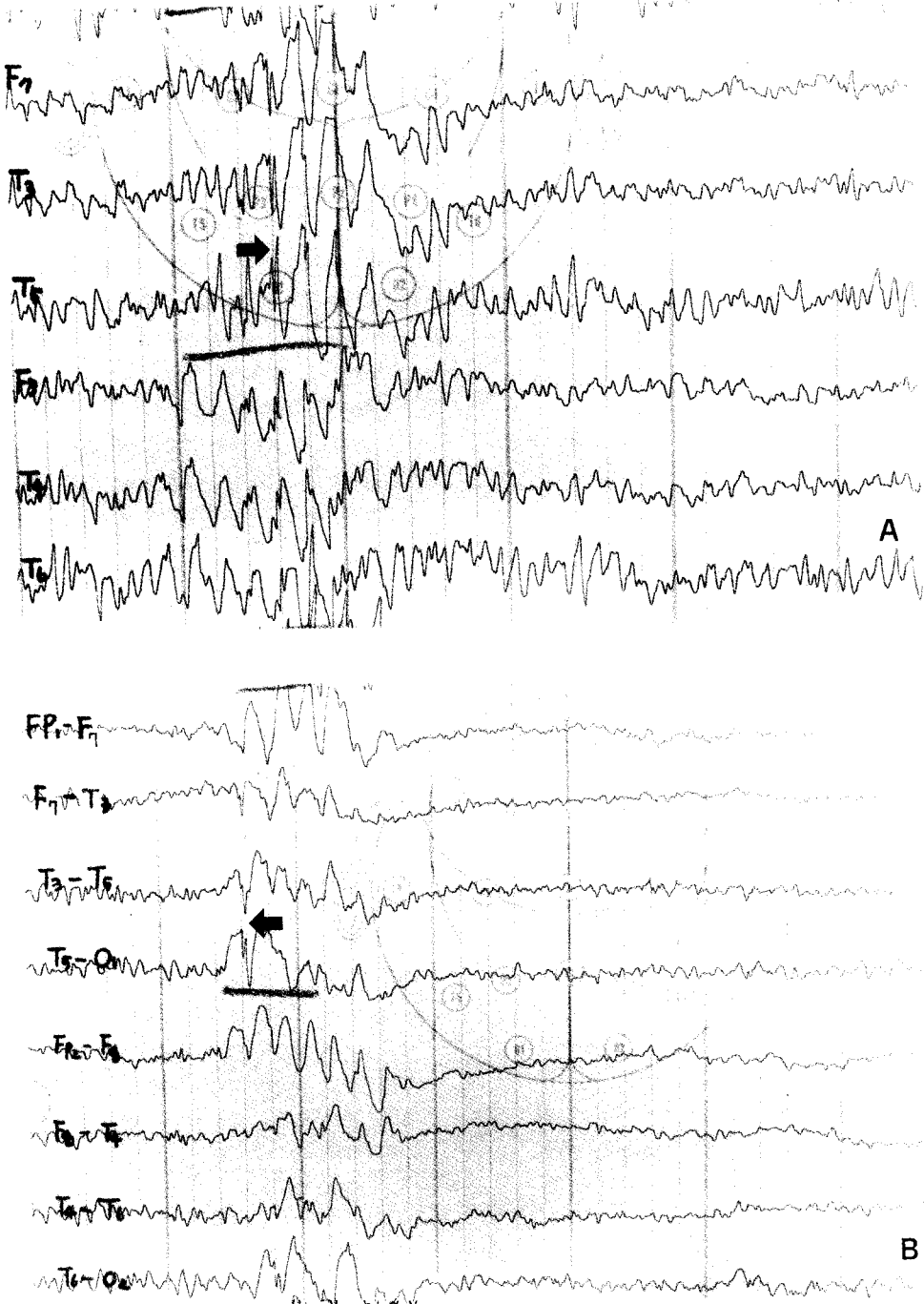


Figure 1. The EEG tracing from case 5(Choi, CB) shows a burst of epileptogenic spikes from the left hemisphere perdominaty(1-A) with phase reversal over the posterior-temporal(T5) region(1-B) (see arrow →)

국문 초록=

성인에서의 반복성, 발작성 복통의 드문 원인 (복부간질)

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성인의 반복성 발작성 복통은 드물게는 측두엽간질에 속하는 “복부간질”로 생길 수 있다. 이 증상은 두통, 안면홍조, 구역질외에 경우에 따라서는 순간적인 의식소실과 같은 중추신경계 자극증상, 간질뇌파형 및 항경련제에 대한 증상치료로 “과민성대장증후군”, “편두통”등 다른 질환과 간별이 될 수 있다. 이번 연구에서 저자는 항경련제 치료로 반복성 발작성 복통이 치료된 다섯명의 환자에서 복부간질로 인한 다양한 위장관질환, 중추신경장애, 특징적인 뇌파소견 및 뇌핵자기공명영상을 검토해 보고자 한다.

Key words : Abdominal epilepsy, recurrent paroxysmal abdominal pain, TLE