A pain fired from brain: Abdominal Epilepsy

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Abstract

Introduction: *Abdominal pain is a nonspecific symptom which can be caused by a constellation of pathologies. It can be related sometimes to pathologies that affect the brain and mind. Among pathological conditions that can cause paroxysmal gastrointestinal symptoms are porphyria, abdominal migraine, cyclical vomiting, intestinal malrotation and peritoneal bands. Among them, rare, but important to recognize is abdominal epilepsy. Abdominal epilepsy is more a diagnosis of exclusion, and it is considered to be part of the group of temporal lobe epilepsies, which usually occur in children. However rare, there is recorded documentation of its occurrence even in adults.*

Method: *We present two adult case reports with abdominal symptoms who have undergone several investigations before the diagnosis.*

Results: *In these patients the pellicular combination of the irregular paroxysm of abdominal pain, combined with symptoms involving the central nervous system and the exclusion of pathologies that can produce similar symptoms can guide the physician to the diagnosis of abdominal epilepsy.*

Discussion: *It should be suspected abdominal epilepsy when we encounter unexplained and recurrent gastrointestinal symptoms such as severe paroxysmal pain, hunger, flatulence, nausea and sometimes diarrhea. Symptoms are associated with central nervous system symptoms such as lethargy, confusion or alteration of consciousness and are improved with antiepileptic therapy. This diagnosis is usually associated with electroencephalography changes. The symptoms are quite vague and for this reason it is difficult to make a diagnosis.*

Keywords: *abdominal epilepsy, unexplained abdominal pain, EEG, adults, anti-epilepsy drug therapy, case report*

Introduction

Abdominal epilepsy (AE) is a rare syndrome in adults that presents with paroxysmal symptoms favoring more an abdominal pathology, but that is fired from the brain in form of seizure activity [(1).] The spectrum of abdominal epilepsy is characterized by: a) periodic abdominal symptoms that can't be explained after extensive medical testing b) symptoms that suggest a central nervous system involvement i.e. confusion, lethargy c) Abnormal electroencephalogram (EEG) with findings for seizure disorder d) sustained absence of abdominal symptoms with antiseizure therapy [(2)]. Zinkin and Peppercorn in their review of Abdominal epilepsy have covered 36 cases reported in literature [(3)]. Due to these nonspecific symptoms and predominance of abdominal symptoms is important to make it known to physicians to avoid misdiagnosing of these symptoms as "psychogenic". [(4)]

Method

Case report

Case 1

Patient 1 is a 40-year-old female. Three years prior to the first assessment at the Neurology Department of University Hospital of Tirana "Mother Theresa" she

experienced a non-convulsive loss of conscience. At the neurological examination it was shown brisk tendon reflexes without other signs. It was recommended to perform brain imaging and EEG, doppler of neck arteries, but the patient refused to undergo these examinations. She did a cardiologic evaluation, which resulted normal.

One year after, the patient had a non-convulsive loss of conscience assumed to be hypoglycemia. After some months she had episodes of recurrent paroxystic abdominal pain, which lasted 5 to 30 minutes, palpitation and flushing of the face, followed by tiredness and lethargy for a few hours.

From several years the patient suffers from headaches with migraines characteristics, related to her menstrual cycle, without aura or focal neurological signs. Headaches had a frequency of four per month and were relieved from non-steroid anti-inflammatory drugs. The headache was further amplified and one month before the second assessment of the patient in the Neurology Department it was severe with an intensity of 10 out of 10, reaching the peak in few minutes, bilateral temporal, throbbing as quality, with photophobia, phonophobia and lasted several hours (more than four hours), without aura or focal neurological symptoms. The pain was resistant to non-steroid anti-inflammatory drugs.

Episodes of abdominal paroxysm were localized at the periumbilical and the left hypochondria with increased frequency and severity during time. The month before the second evaluation they become daily (several episodes per day) with an urge to defecate. After that the patient had lethargy and feeling "dull" for several minutes to hours. Sometimes they were associated with headaches.

Regarding personal medical history: She has alopecia. The patient didn't undergo surgeries and do not use any medications (except the ones for headache attacks), doesn't use alcohol or illicit drugs, and doesn't smoke.

Blood and biochemical test were unremarkable. TSH, Vitamin B12 and Vitamin D showed at the normal range. The patient carried out several investigations: cardiology investigation, chest radiography, endocrinology and rheumatology investigation which resulted normal. Gastroenterology and infectious disease investigations also resulted unremarkable. ECHO Doppler of the neck arteries and temporal arteries showed normal results.

We carried out the tests for ANA; Anti DNA; Antiphospholipid antibodies; Anca-c; Anca- p which resulted negative. Lab results for Homocysteine; Protein S; Protein C; Antithrombin III; Factor V (Leiden): ACE also showed normal results.

FIGURE 1: MRI of the head: Sucentrimetric lesions of periventricular subcortical white matter and bilateral subcortical areas of cerebral hemispheres. Without enhancement after gadolinium.

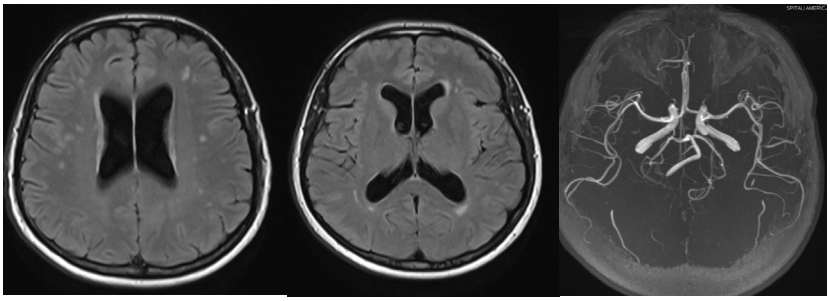


FIGURE 2: MRI of spine: Paracentral protrusion of C4-C5 intervertebral disc. Left foraminal protrusion of C5-C6 intervertebral disc. Central protrusion of the Th6-Th7 intervertebral disc.

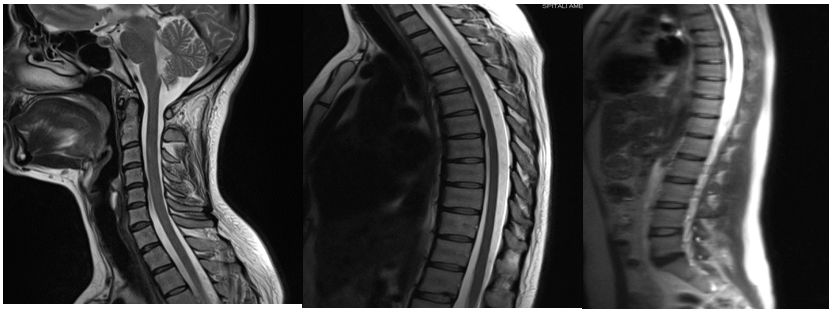


FIGURE 3: EEG: Slow waves with high voltage at the frontal regions more evident in the left derivations

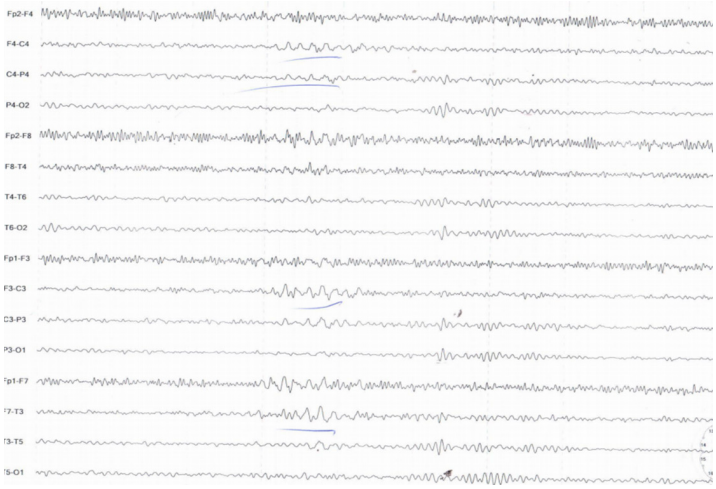
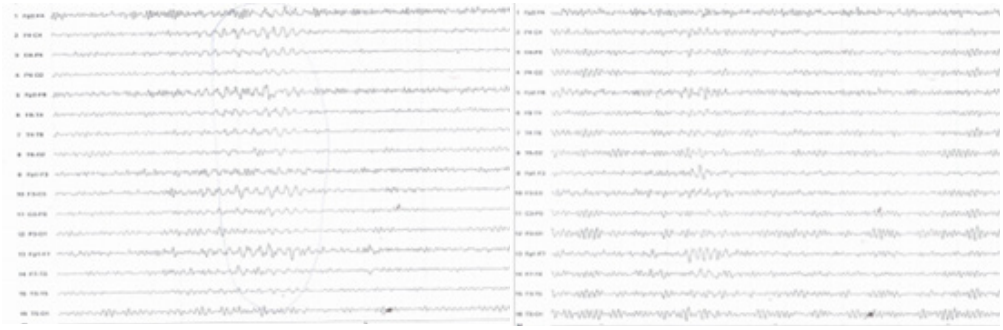


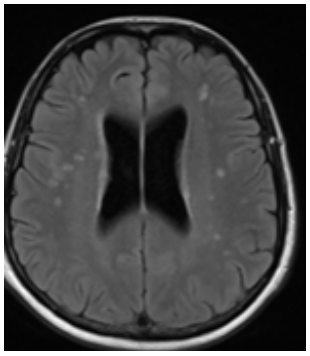
FIGURE 4: EEG (sleep deprivation): Slow waves with high voltage at the frontal regions more evident in the left derivations



The patient was previously assessed by a psychiatrist and he recommended to take SSRI. The episodes became more often during the treatment with SSRI; Also, it was referred that one episode was associated with twitching for few minutes of the right leg (clonic) and loss of conscience.

After the neurological consultation the SSRI was interrupted and it was started the antiepileptic therapy with Levatiracetam 1500 mg/daily. Over the course of the disease the symptoms improved, and the patient was seizure free after four months. She experiences headaches rarely.

FIGURE 5: MRI of the head at follow up (after two years)



MRI of the head after two years was without changes. The patient is still seizure free; we followed her up for several years and the therapy with levetiracetam slowly tapered. The patient rarely experiences headaches, with a frequency once per month.

Case 2

Patient 2 is a female, 33 years old, with a history of two episodes of syncope in ten years. Since several months the patient suffered from episodes of severe abdominal pain, paroxistic and severe, urge to defecate, pallor, sweating, without palpitations. Several minutes after the episodes the patient referred alteration of awareness and difficulty to speak. The duration of the episode was approximately 50 minutes, one episode was accompanied with loss of consciousness (non-convulsive). Episodes were rare at the beginning (once in several months/once a year) and were increased during the last year with a frequency of once in three months. One month before the appointment the patient had a loss of consciousness.

After the episode she had moderate headache at the occipital region, without nausea and vomiting, without photo-phono phobia or any neurological sign. The headache lasted several hours and was relieved by paracetamol and other non-inflammatory steroid drugs. The frequency of headaches was about one in three days. The patient is the fourth child in the family. The development and milestones were met normally. The patient had no febrile seizures. She referred to an episode of head trauma at the age of seven years old with loss of conscience. The patient also suffers from rheumatoid arthritis for several years and is under treatment with Medrol, Methotrexate, Ipsoflog. The patient doesn't use alcohol, illicit drugs, or smoke. There are no diagnosed neurological diseases in her family. Her mother suffers from dermatitis.

Blood and biochemical tests were carried out, thyroid tests and vitamins levels showed normal results. Cardiology evaluation and gastroenterology evaluation were carried out and the findings were unremarkable. Chest radiography was normal. CT of the head was done after the first episode with loss of conscience. Thereafter was performed a thoraco-abdominal CT with contrast which showed unremarkable findings.

FIGURE 6: MRI of the head: Two subcortical nonspecific lesions

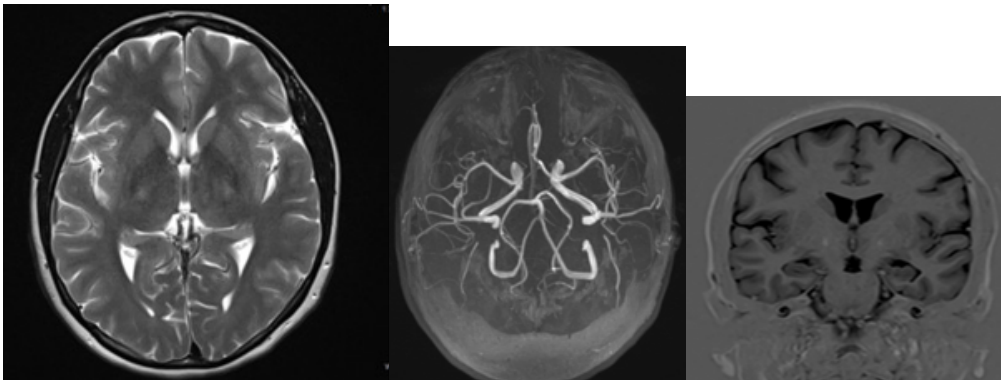
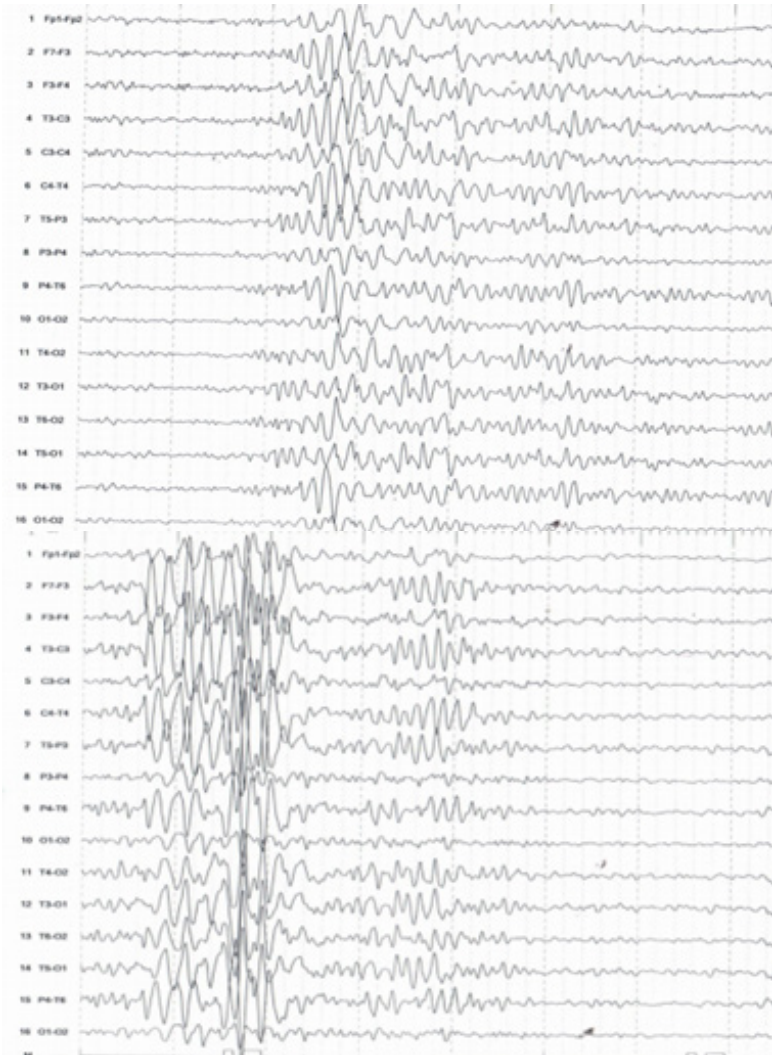
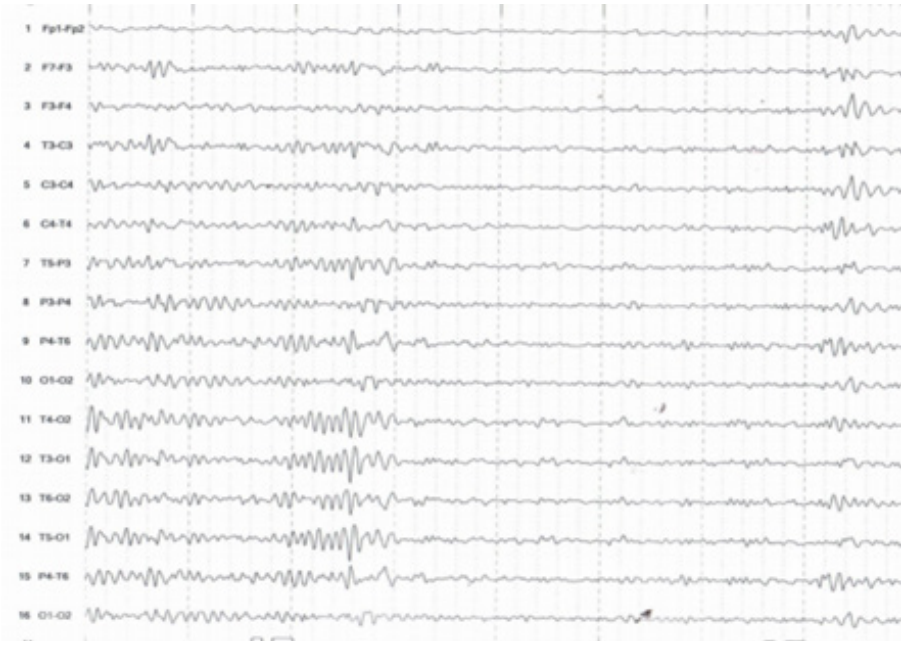


FIGURE 7: EEG: Diffuse paroxistic discharges



It was started the medication with anti-epileptic drugs Levatiracetam (1000 mg/day). The patient was seizure free for four months. During the follow up in seven years she had one episode in a year.

FIGURE 8: EEG after one month from the medication (improved)



Discussion

Abdominal epilepsy is a rare disorder, and it is often a missed diagnosis. It commonly occurs in the pediatric age group, but there is also documentation of its occurrence in adults [5]. It is characterized by otherwise unexplained, paroxysmal gastrointestinal symptoms and co-occurrence of central nervous system symptoms and abnormal EEG findings that point to a seizure disorder and are improved with antiepileptic drug therapy [3].

Abdominal epilepsy has multiple presenting symptoms. Gastrointestinal symptoms include abdominal pain presented more like short lasting cramping, nausea, hunger, flatulence, and diarrhea. The central nervous system symptoms are dizziness, lethargy, headache, mostly migraine like, confusion, loss of consciousness, and transient blindness [6]. Abdominal epilepsy is a type of autonomic epilepsy that can be associated with autonomic symptoms as sweating, pallor or cold. [7]

Although the several hypotheses, the cause of abdominal epilepsy remains still unclear. Some hypotheses relate the sylvian fissure and insular cortex as the origin of the seizure since they coincide with the locations of the abdomen on the sensory homunculus. Moreover, the portion M2 of MCA (middle cerebral artery) goes through the sylvian fissure [8]. Any pathology of the vessel, particularly at this specific segment, could be assumed to play a role in seizure activity arising from

the temporoparietal lobes. There are some reported cases of ictal abdominal pain associated with right parieto-occipital encephalomalacia, bilateral parietal atrophy and bilateral perisylvian polymicrogyria [9].

Recurrent abdominal pain is seen in abdominal migraine, peptic ulcer, and visceral hyperalgesia[10]. Abdominal epilepsy has overlapping features mostly with abdominal migraine, but the duration of symptoms is longer in abdominal migraine and EEG is usually without seizure activity, contrarily to abdominal epilepsy [6].

Conclusions

These cases show how challenging and time-consuming is the diagnosis of abdominal epilepsy, especially in adults. We need to keep in mind that abdominal epilepsy is a diagnosis that could often be missed by physicians. It should be taken as possibility in patients who present with episodic, recurrent, and paroxysmal gastrointestinal complaints along with symptoms which suggest the central nervous system involvement, after being excluded the most common causes and the symptoms do not improve with standard treatments. Also, we must be aware of misdiagnosis, especially for women who present with complains which do not fit in common diagnoses. When other diagnoses are excluded with routine examination, we should be cautious to not give the diagnosis of a somatoform disorder. In these cases, it is important to know and see the repeated pattern and we recommend considering an EEG (or a video-EEG) examination before labelling these symptoms as "psychogenic".

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