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When the Stomach Pain Is Literally "In Your Head"

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Summary. Abdominal epilepsy (AE) is an extremely rare condition, classified as temporal lobe epilepsy, and is usually a diagnosis of exclusion. Temporal lobe epilepsy often has no clear cause, although it may be associated with diseases such as temporal lobe sclerosis, dysembryoplastic neuroepithelial tumours, and other benign tumours, as well as arterio-venous malformations, gliomas, defects in neuronal migration, or lesions of the cortex caused by encephalitis. AE is more common in children but has been also reported in adults. AEs are characterised by recurrent and unexplained gastrointestinal symptoms such as seizure pain, nausea, bloating and diarrhoea, which improve with antiepileptic treatment. Given the vague nature of these symptoms, patients are at high risk of misdiagnosis. An electroencephalogram and neuroimaging of the brain are needed to confirm the diagnosis.

We present the clinical case of a 67-year-old female patient who was investigated at the Gastroenterology Department for a sharp pain in the left side of the abdomen, frequent abdominal distension and gurgles, diarrhoeal episodes, weight loss, paroxysmal hallucinations, and headaches. After a thorough gastroenterological examination, consultations with a psychiatrist and a neurologist, an MRI and an EEG were performed and the patient was diagnosed with focal temporal lobe epilepsy.

Keywords: abdominal epilepsy, temporal lobe seizures, gastrointestinal symptoms, hallucinations.

INTRODUCTION

Abdominal pain is a non-specific symptom that can be caused by a variety of underlying diseases and is often misdiagnosed [1]. As abdominal epilepsy (AE) is considered to be an unlikely cause of this symptom, it may be overlooked in the differential diagnosis, as there are many more common conditions associated with paroxysmal abdominal pain [2]. Other pathological conditions, such as biliary colic, cyclic vomiting, visceral hyperalgesia, abdominal migraine, or peptic ulcer disease, may also present with paroxysmal gastrointestinal symptoms [3, 4]. AEs are characterized by paroxysmal-epizodic abdominal and perineal pain caused by central nervous system disturbances [5]. As in other epilepsies, specific abnormalities in electroencephalogram (EEG), as well as episodes of impaired awareness, post-episode somnolence, and a favorable response to anti-seizure medications can be seen in these cases [6, 7]. In addition, emotional and psychological factors may contribute to the manifestation of gastrointestinal disorders in some patients, making accurate diagnosis difficult and sometimes attributing symptoms to "functional" or "psychogenic" causes [8]. Due to varying symptoms, patients often seek medical assistance from general practitioners, surgeons, gastroenterologists, or psychiatrists, with fewer referrals to neurologists. In some cases, patients undergo an exploratory laparotomy unnecessarily in the absence of significant symptoms [2]. Therefore, it is very important to distinguish AE from other gastrointestinal pathologies to avoid unnecessary procedures and to alleviate prolonged patient suffering. In this paper, we present a rare clinical case of a 67-year-old female patient with abdominal epilepsy caused by a tumourous mass in the mesiotemporal lobe. The patient could have been misdiagnosed as having CCC with biliary colic attacks, potentially leading to the removal of her gallbladder, as AE and CCC share certain clinical similarities. However, the patient also presented with various symptoms of psychiatric disorders, including anxiety and depression, which had been treated for an extended period without considering the origin of the symptoms.

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CASE REPORT

A 67-year-old female patient presented to the Gastroenterology Department complaining of sharp abdominal pain on the left side and in the left groin area, frequent abdominal bloating and gurgles, frequent bowel movements, weight loss, recurrent hallucinatory episodes, and headaches. She had also noticed fresh blood in her stool. Objective examination revealed an asthenic physique. Upon examination, the patient complained of palpation tenderness in the left groin area. A complete blood count performed on the day of hospitalization showed no abnormalities. The patient did not present with jaundice. She denied any chronic diseases or harmful habits.

According to the patient's detailed medical history, these gastrointestinal symptoms had appeared approximately a year earlier, after a cholecystectomy procedure had been performed due to chronic calculous cholecystitis (CCC). The patient had lost 15 kg of weight since then.

Seven years ago, the patient underwent surgery for cervical cancer, after which she developed anxiety, episodic hallucinations, and depression. All these symptoms worsened after cholecystectomy with no gastrointestinal symptoms. For bad psychiatric conditions, the patient was treated twice in the psychiatric ward with mirtazapine and lorazepam without significant improvement. Two months before recent hospitalization, the patient underwent a video colonoscopy, which revealed radiation proctopathy without any signs of bleeding.

In the Gastroenterology Department, the abdominal ultrasound showed only minor fibrotic changes in the liver. The computer tomography (CT) scan revealed arterial phase enhancement in the peripheral part of segment 4a of the liver, a mid-third segment aneurysm of the splenic artery, and incidental findings in the pelvis. A chest X-ray was also performed, which showed no significant abnormalities. Negative HBV, HCV infection markers and autoantibody tests against liver antigens helped to rule out infectious and autoimmune hepatitis.

Overall, no somatic gastrointestinal pathologies were found and she was referred for psychiatric consultation. It became clear, that all 'hallucinations' were accompanied by epigastric discomfort, derealisation, and amnesia after the episode. They lasted up to 2 minutes, ended spontaneously or after sedatives, and recurred several times a week. A neurological consultation was therefore recommended. The patient also recalled that five years ago, she had experienced a loss of consciousness (LOC) with generalized jerks, which had led to an evaluation at the emergency department, including a CT scan. However, no evidence of brain abnormalities had been detected. The second LOC appeared two years later, but epilepsy was not suspected and consultation with an epileptologist was not requested.

Brain magnetic resonance imaging (MRI) and electroencephalography (EEG) were requested. MRI showed an increased signal and volume of the left uncus, hippocampal crus, dentate gyrus, and amygdala (Fig. 1). An EEG revealed localized epileptiform potentials in both temporal regions with a clear emphasis on the left side (Fig. 2). The patient was diagnosed with focal epilepsy with frequent focal and occasional generalized convulsive seizures, and carbamazepine 200 mg twice daily was initiated.

Three years after the initial diagnosis, the patient had a follow-up consultation with an epileptologist, and, according to her, she no longer experienced seizures. Follow-up brain MRI showed decreasing changes in the anterior part of the left mesiotemporal region (Fig. 3) and EEG showed improvement with accidental epileptiform potentials in the left temporal region (Fig. 4).

DISCUSSION

We present a case of a patient with epilepsy who had been investigated and treated by gastroenterologists and psychiatrists for a number of years for non-specific complaints. The diagnosis was complicated by the fact that the patient herself referred to her epileptic seizures as hallucinations and always emphasised only the paroxysmal abdominal pains. Someone once described/suggested the term "abdominal epilepsy" [2]. This is a rare disorder characterized by vague symptoms, often leading to misdiagnosis or missed diagnosis. It is typically categorized as a form of temporal lobe epilepsy and frequently considered a diagnosis of exclusion [9]. While AE is typically observed in the pediatric age group, there have been documented cases of its occurrence in adults as well [9]. In our patient's case, the onset of AE was unique, as it began at the age of 60. Among the rare forms of epilepsy, AE stands out. It is characterized by sudden-onset paroxysmal pain in the midline or upper abdomen, which lasts for a few minutes or less. Other symptoms, such as nausea, vomiting, headaches, and neurological complications, may also be present [10]. In our patient's case, the paroxysmal abdominal pain was localized in the left upper and lower quadrants, along with bloating, which has also been observed in other AE patients [5]. Notably, our patient exhibited not only an AE expression but a charade of psychiatric symptoms also, persisting for many years. This combination of gastrointestinal (GI) and mental manifestations raises concerns about possible structural alteration in the mesiotemporal lobe, such as DNETs, amygdala body dysplasia, or hamartoma [2]. In medical cases involving such abnormalities in the temporal focal regions, concurrent limbic or experiential auras such as fear, déjà vu, derealization, and epigastric sensations have been reported as primary symptoms. These psychiatric symptoms tend to worsen during and after a seizure.

In order to exclude other, usually more common causes, a comprehensive evaluation should be conducted, including physical and neurological examinations, laboratory studies, endoscopy, and abdominal imaging. Computed tomography (CT) and ultrasound are typically used for abdominal imaging [11]. In the case of our patient, she presented with a tumorous-like lesion in the mesiotemporal lobe, raising suspicion of a DNET. MRI would also be beneficial to further investigate likewise cases [12].

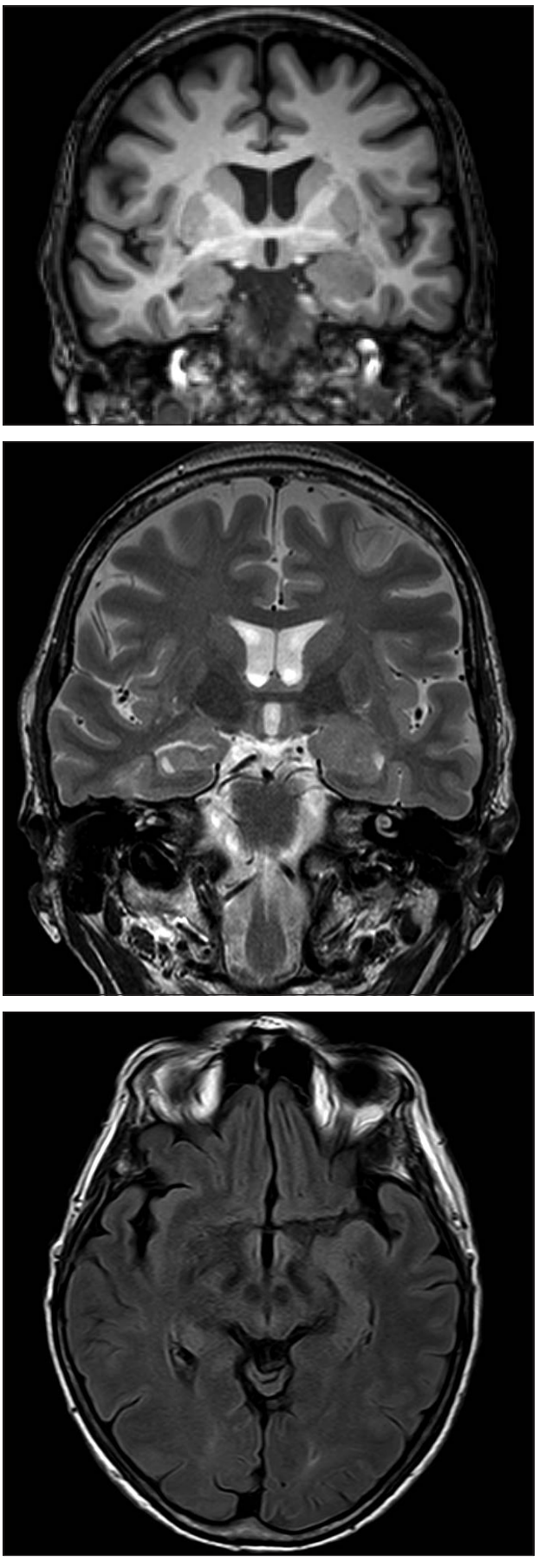

Fig. 1. **Brain MRI showed an increased signal and volume of the left uncus, hippocampal crus, dentate gyrus, and amygdala**

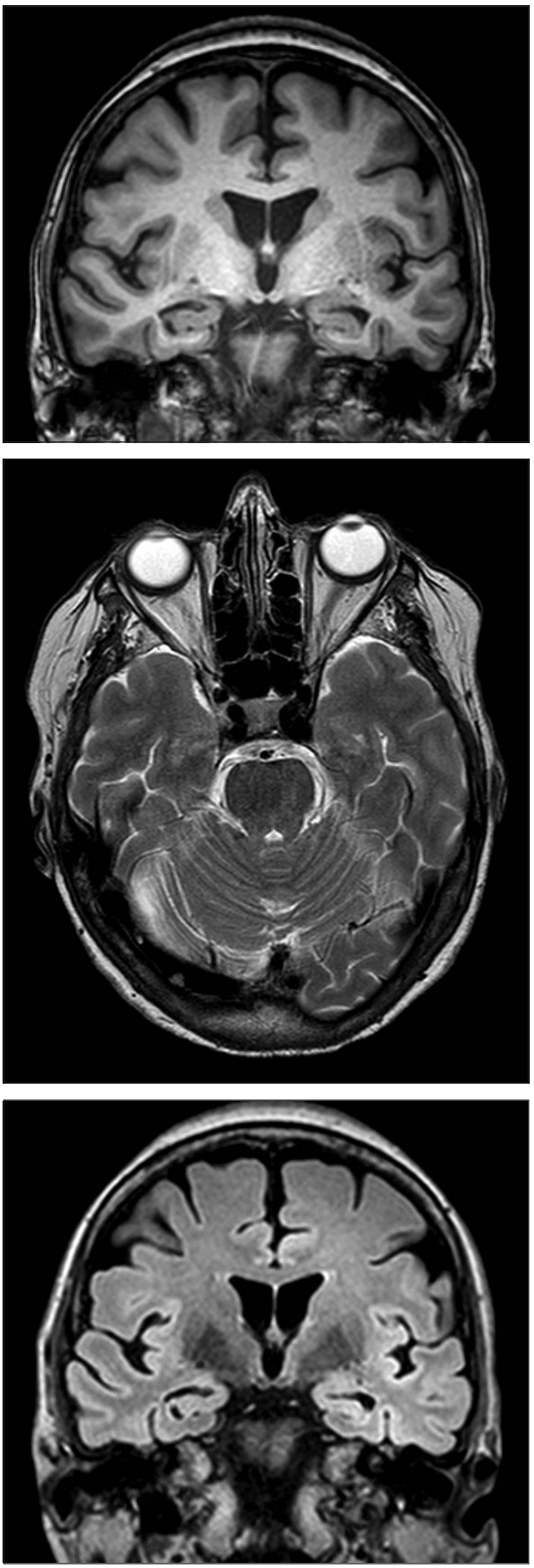

Fig. 3. **Follow-up brain MRI**

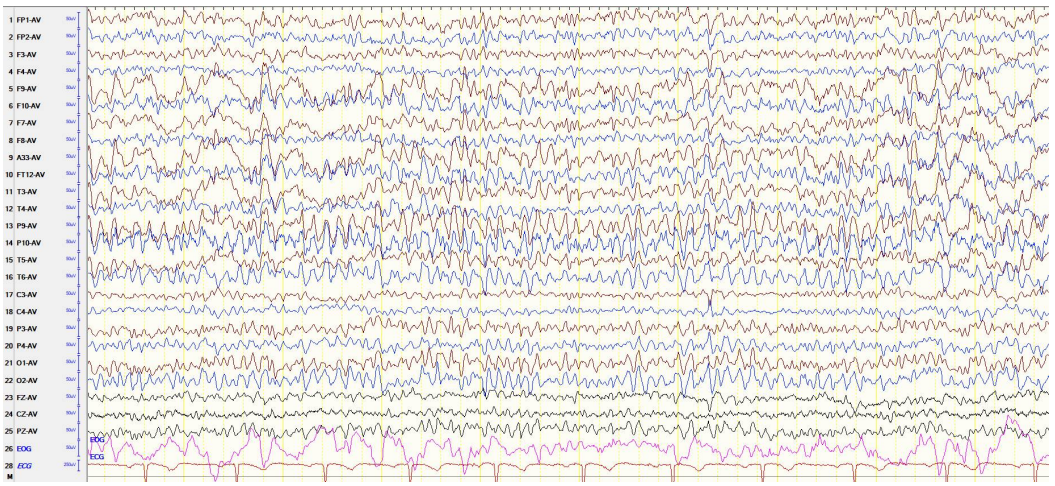

Fig. 2. **EEG. Localized epileptiform potentials in both temporal regions with a clear emphasis on the left side**

The 4 main diagnostic criteria for AE include:

1. Otherwise unexplained, paroxysmal GI complaints, mainly pain and vomiting;

2. Symptoms that arise from a central nervous system disturbance;

3. Abnormal EEG with findings specific to a seizure disorder (reports have indicated that the most frequent EEG findings in patients with abdominal epilepsy are sharp waves and/or spikes originating from one or both temporal regions [7, 13];

4. Improvement with anticonvulsant medication (in the case of our patient, carbamazepine was prescribed at a dosage of 200 mg twice daily, which resulted in the patient becoming seizure-free);

Our patient fulfilled all four diagnostic criteria.

The exact underlying mechanism of focal epilepsy with ictal abdominal pain remains unknown. However, studies have suggested that certain brain regions, such as the Sylvian fissure, anterior insular cortex, frontal operculum, and mesiotemporal structures, may play a crucial role in explaining the origin of this condition [6, 14]. It is thought that the amygdala, a part of the brain associated with emotional processing, may contribute to the manifestation of GI symptoms, as well as generalized anxiety. The amygdala relays signals to the GI tract through direct projections to the dorsal motor nucleus of the vagus nerve. Additionally, the hypothalamus is believed to activate sympathetic pathways from the amygdala to the gastrointestinal tract, further contributing to the manifestation of GI symptoms [15].

Considering that our patient's symptoms were attributed to a likely DNET, the electrical activity could be explained by two approaches: the tumor-centric, and the epilepsy-centric [16]. The tumor-centric theory suggests that the epileptic activity originates directly from the tumor itself. On the other hand, the epilepsy-centric hypothesis emphasizes the role of metabolic imbalances associated with glioma-related changes in glutamatergic and gamma-aminobutyric acid (GABA) signalling that contribute to epileptogenicity in tumor-related epileptic activity [17].

Once the diagnosis of AE is confirmed, treatment can begin, following similar approaches as other forms of epilepsy. Typically, treatment commences with the administration of anti-seizure medications (ASM). In our patient's case, treatment with carbamazepine effectively eliminated AE attacks. However, in cases where pharmacotherapy proves ineffective, appropriate neurosurgery can offer a potential cure or, at the very least, make the condition more manageable with medication [2].

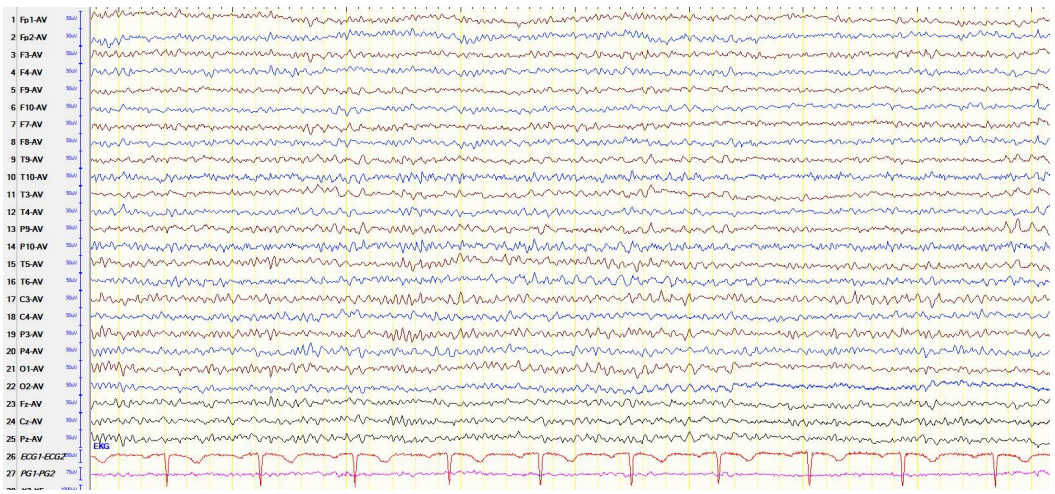

Fig. 4. **Follow-up EEG**

DIFFERENTIAL DIAGNOSIS

Chronic calculous cholecystitis (CCC). It is important to mention biliary colic, as our patient had a gall bladder removal due to CCC one year before her visit to the Vilnius University Hospital Santaros Klinikos. This could have been a mistake made by clinicians in misinterpreting biliary colic with AE episodes. CCC refers to a long-lasting gallbladder inflammation primarily caused by gallstones obstructing the cystic duct [18]. While both conditions may cause recurrent or persistent abdominal pain, their underlying causes, diagnostic criteria, and treatment approaches differ significantly. The first thing to recognize when differentiating both conditions is the duration of pain and its characteristics. CCC manifests with recurrent or persistent abdominal pain, often located in the right upper quadrant. The duration of pain episodes can range from minutes to hours, lasting for several days in some exceptional cases. If CCC presents with a biliary colic, the pain usually lasts 30 minutes to several hours with a cramping sensation [18]. The abdominal pain in AE is typically paroxysmal, lasting up to 2 minutes, and, most importantly, is accompanied by altered consciousness. Our patient had also lost consciousness during a seizure episode, as she could not clearly remember what happened during an AE episode. In both CCC and biliary colic, the pain lasts up to 2 minutes and consciousness does not tend to be affected. Also, CCC has some triggers that provoke pain, such as fatty meals, whereas regarding AE there are no specific triggers [18]. Diagnostically, CCC involves the gallbladder and requires imaging studies, such as abdominal ultrasound, while AE is a neurological disorder diagnosed through EEG and is treated using ASM.

Abdominal migraine is the most common condition that shares similarities with AE, making it challenging to differentiate between the two, especially when patients present with abdominal symptoms and headaches. Our patient also experienced a predominantly left-sided headache. Abdominal migraine is characterized by recurring episodes of abdominal pain, primarily around the belly button or with poorly localized pain. These episodes are often accompanied by other migraine-related symptoms, such as nausea, vomiting, and sensitivity to light and sound, that last hours to days [19]. Whereas, in our patient, the pain episodes were typically brief, lasting a few minutes, and occurred alongside neurological symptoms like altered consciousness and other manifestations related to seizures. A family history of migraine is often present [20]. Moreover, EEG abnormalities consistent with epilepsy and a positive response to anticonvulsant therapy are indications for AE, but these do not correlate with abdominal migraines.

Visceral hyperalgesia (VH) refers to heightened sensitivity and exaggerated pain perception in response to normal or slightly noxious stimuli within the internal organs. While VH and AE can present with abdominal pain, VH is more commonly associated with functional gastrointestinal disorders, whereas AE is a specific form of epilepsy with abnormal brain activity. The pain is usually associated with eating: it can occur after consuming food, following a relief after eating, or starving for a while. However, our patient did not mention the relation of food ingestion with reoccurring abdominal pain. It is not uncommon for patients with visceral hyperalgesia to present with headaches, increased sweating, muscle tension, irritable bladder, or irritable bowel syndrome. Our patient suffered from frequent headaches, although the other symptoms mentioned could also have occurred, since the patient had anxiety, which go concurrently with muscle tension and other VH symptoms mentioned. The pain can be diffuse or localized to specific areas. VH also has triggers, such as stress, infections, certain foods, and emotional excitements, whereas AE has no specific triggers [21]. AE differentiates from VH by specific EEG and MRI findings. VH is treated with proton pump inhibitors and, in some cases, antidepressants, while AE is treated with anti-epileptic drugs [21].

Peptic ulcer disease. As for peptic ulcer disease, the clinic is similar to one of VH. The pain usually occurs in the epigastrium, can irradiate to the back, and is associated with increased hydrochloric acid secretion. The pain is also associated with food as it occurs shortly after eating in stomach ulcers and 2-5 hours after eating in duodenal ulcers [22]. As mentioned above, our patient had paroxysmal abdominal pain that lasted only few minutes and was not associated with food ingestion. In addition, peptic ulcers can bleed, thus melena can be present. Melena is defined as black tarry stool with a glistering sheen and results from degradation of blood in the GI tract. There were traces of fresh blood in our patient's stool, which usually appear from bleeding areas in the lower GI tract, most likely the rectum. Melena prevails mostly from upper GI tract bleeding zones, that is, the stomach.

CONCLUSION

The case described illustrates the difficulties and time taken to make a definitive diagnosis of AE, primarily due to the vague symptomatology and low suspicion, particularly in adults. Given the potential for AE to be overlooked or misdiagnosed, it is crucial to consider this condition in patients experiencing episodic, recurrent, and paroxysmal GI complaints accompanied by symptoms indicative of CNS dysfunction that do not respond to standard treatments. Otherwise, as mentioned above, patients may undergo unnecessary procedures or even operations. Moreover, in such cases. it is beneficial to acknowledge the stigma associated with somatoform disorders and promptly consider an EEG to prevent mislabeling symptoms as "psychogenic" or "functional". If abnormal EEG findings are present, treatment involves the initiation of anti-epileptic drugs with regular follow-up. In the case of our patient, the administration of 200 mg carbamazepine twice daily proved highly effective in managing AE.

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KAI PILVO SKAUSMAS YRA GALVOJE

Santrauka

Abdominalinė epilepsija (AE) yra ypač reta temporalinės skilties epilepsijos rūšis. Ji dažniau pasireiškia nesant struktūrinio smegenų pažeidimo, tačiau priežastis gali būti mezialinė temporalinės skilties sklerozė, disembriogeniniai neuroepiteliniai tumorai ir kiti gerybiniai navikai, arterioveninės malformacijos, gliomos, neuronų migracijos defektai ar gliozės po persirgto encefalito, traumos ar kraujotakos sutrikimų. Nors AE dažniausiai pasireiškia vaikams, yra pasitaikę atvejų ir suaugusiesiems. Klinikai būdingi pasikartojantys ir nepaaiškinami gastrointestinaliniai simptomai, tokie kaip paroksizminis pilvo skausmas, pykinimas, pilvo pūtimas, viduriavimas, sumažėjantys pradėjus vartoti prieštraukulinius vaistus. Greta neretai pasireiškia ir suvokimo sutrikimai, mieguistumas po priepuolio. Atsižvelgiant į neaiškų šių simptomų pobūdį, susidaro didelė klaidingos diagnozės tikimybė. Ligos diagnostikai ir kontrolei yra svarbus elektroencefalogramos tyrimas ir teigiamas atsakas į prieštraukulinius vaistus.

Šiame straipsnyje pristatome 67 metų pacientės, daugelį metų gydytos gastroenterologų ir psichiatrų, atvejį. Moters liga manifestavo aštraus pobūdžio skausmais kairėje pilvo pusėje, dažnu pilvo pūtimu ir gurguliavimu, skystomis išmatomis, svorio netekimu, haliucinacijų epizodais ir galvos skausmais. Po vizitų pas įvairius specialistus ir atliktų magnetinio rezonanso tomografijos (MRT) ir elektroencefalografijos (EEG) tyrimų, pacientei buvo diagnozuota židininė temporalinė epilepsija, o skyrus gydymą karbamazepinu, išnyko visi gastroenterologiniai ir psichiatriniai simptomai.

Raktažodžiai: abdominalinė epilepsija, temporalinės skilties epilepsija, klinikinė charakteristika, požymiai.

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