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EDITORIAL

Abdominal Epilepsy, a Rare Cause of Abdominal Pain: The Need to Investigate Thoroughly as Opposed to Making Rapid Attributions of Psychogenic Causality

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Introduction

Abdominal pain is a nonspecific symptom which can be caused by myriad pathologies, resulting in frequent misdiagnosis.[1] Some pathological conditions can cause paroxysmal gastrointestinal symptoms, such as porphyria, cyclical vomiting, intestinal malrotation, peritoneal bands, and abdominal migraine.[2] Furthermore, emotional and psychological factors may also play an important role in the presentation of certain patients with gastrointestinal disorders, and accurate diagnosis can be confounded by these. An accurate diagnosis may be delayed or even abandoned due to the attribution of "functional" or "psychogenic" causality.[3] Physicians in numerous fields of practice too often respond in such a fashion when the more common causes of pain conditions are ruled out,[4] which potentially puts patients with rare pain disorders that are challenging to diagnose at considerable risk for needless, prolonged suffering. Further, the stigma associated with being diagnosed with a Somatoform Disorder or a Medically Unexplained Symptom (MUS) should not be understated.[5]

One extremely rare cause of abdominal pain is abdominal epilepsy, also known as autonomic epilepsy.[6] The typical symptoms are idiopathic, paroxysmal-episodic abdominal and periumbilical pain caused by a central nervous system disturbance.[7,8] In addition, as with common epileptic disorders, abnormal electroencephalograms (EEGs) are usually observed (assuming they are performed), as well as loss or alteration of consciousness, somnolence following episodes, and a favorable response to antiepileptic drugs.[6,9,10] In some patients, the irregular bouts of abdominal pain, combined with the above symptoms, can point the physician towards a diagnosis of abdominal epilepsy.[2]

Case Study

To illustrate, we present the case of a fifty-two-year-old woman with a 15-year history of recurrent, paroxysmal abdominal pain, absence episodes and two genralized seizures diagnosed as abdominal epilepsy by a gastroenterologist and neurologist following a lengthy period of clinical assessment.

The abdominal epilepsy was experienced as abdominal pain occurring suddenly, resolving spontaneously and lasting for 10–15 mins with palpitation and stuttering. These attacks occcured at random intervals, sometimes every few days and

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sometimes once a month. Her symptoms during these episodes included cramps, episodic vomiting, headache and pain experienced as a pressure sensation in her abdomen that radiated to her lumbar area. Although the patient experienced two episodes of generalized seizures, her abdominal pain was typically not associated with convulsions or loss of consciousness, although was typically followed by somnolence, lethargy, and increased sleep. The patient also reported absence episodes, independent of the abdominal pain. She described the absence episodes as "like watching the world from the TV—I know what is happening but I cannot interact." The patient's pain was reportedly not helped by any medication.

At the age of 10 years, the patient reportedly experienced an episode in which she felt dizzy and lost the ability to concentrate, experienced frequent odd sensations in her left ear persisting until the present and, occasionally, a pulling sensation on her teeth. In spite of the unremarkable results yielded by an EEG investigation, the episodes of compromised consciousness continued. Her emotional response to these symptoms was a depressed mood throughout her adolescence and she was consequently referred for counseling. During her mid-20s, the patient continued to experience sensations in her left ear, with these symptoms progressing to the point that she believed that she was suffering from some type of auditory condition. At the age of thirty seven, the patient suffered her first episode of the abdominal symptoms combined with a generalized seizure. Specifically, she awakened with abdominal pain during the night and went to the toilet, where she collapsed and had a generalized seizure. This event included tongue biting and incontinence, with the entire episode lasting ten minutes. She was confused postictally, and the patient required several days to fully recover. She was prescribed carbamazepine and reportedly did not suffer any further generalized seizures for well over a decade.

At the age of fifty, the patient suffered another generalized seizure. As was the case in her initial seizure, she reportedly experienced abdominal pain immediately prior to the generalized seizure. Symptoms included loss of consciousness and involuntarily shaking of her limbs, although no incontinence or tongue biting. Following this second seizure in 2018, the patient underwent an MRI of her brain. Results indicated the presence of a pituitary lesion which was further evaluated with dedicated pituitary imaging. This imaging confirmed a 10 mm-sized Rathke's cyst. Although an endocrinological referral was made, all endocrinologic assessments were within normal limits.

At that point, the patient underwent exhaustive investigations including routine blood tests, electrocardiography, abdominal ultrasound and upper gastrointestinal endoscopy showing only a hiatal hernia. Gastric biopsies were normal and no other abnormalities were found in any of the other general medical and neurological investigations. An EEG record was within normal limits.

Adding pregabalin reduced the frequency and severity of abdominal pain and associated lethargic episodes. The patient's current medication regimen is fluoxetine 20 mg bid, carbamazepine 200 mg qam and 400 mg qhs, pregabalin 75 mg bid, and codeine/paracetamol 30 mg/500 mg up to eight tablets qd prn. The patient denies any subsequent seizure episodes, and reports abdominal pain control with which she is satisfied. According to the patient, the addition of the pregabalin was the factor that she saw as most salient to her recovery.

Written consent to publish this report was provided from the patient prior to authors' initiation of the writing of this article.

Discussion

The pathophysiology and etiology of abdominal epilepsy remain unknown. Research suggests that the insula and Sylvian fissure might play an important role, contributing to the etiology of the pathology.[10] In some cases, brain tumors have been assessed as the cause of abdominal epilepsy.[6,11] In other investigations, the possible role of somatosensory area I of the brain in pain perception has been suggested, with the development of partial seizures representing a potential link between abdominal pain and a parietal lobe hemorrhage.[12] Other studies on abdominal epilepsy have indicated that linked causes could be right parietal and occipital encephalomalacia, biparietal atrophy and bilateral perisylvian polymicrogyria.[13] In addition to abdominal pain, the main symptoms associated with focal epilepsy with ictal abdominal pain or abdominal epilepsy include nausea, vomiting, lethargy, and hunger.[10,14] A 2001 study demonstrated that over 4% of patients with focal epilepsy experienced painful epileptic auras, with 5% experiencing abdominal pain in cases of temporal lobe epilepsy and 50% experiencing abdominal pain among those with frontal lobe epilepsy.[15] Numerous studies have suggested that in some cases, abdominal symptoms might be the only element of epileptic activity.[2,10,16,17]

The key features required in specifying a diagnosis of abdominal epilepsy are often idiopathic and irregular abdominal symptoms, loss of consciousness and focal

abnormal EEGs,[6,10,15] although not all of these symptoms need to be present in order to make such a diagnosis. Most patients diagnosed with abdominal epilepsy have registered abnormalities in their EEGs, such as high vol tage, slow waves and generalized spike and wave discharges.[2,6–11,14–16] Furthermore, it is important to note that when a patient experiences an attack, multiple symptoms can manifest concomitantly, including nausea, headache, loss of consciousness and hallucinations.[16] Early diagnosis of abdominal epilepsy can be achieved if it is noted that the attacks occur suddenly, are of brief duration, and resolve spontaneously. Furthermore, the localization of the pain is important to note, as it is typically found in the periumbilical or upper abdominal areas.[2,16] The patient in the case study we have presented experienced repeated attacks of severe abdominal pain as part of the manifestation of epileptic seizures over fifteen years. Abdominal epilepsy remains a rare condition and should only be considered if there are idiopathic paroxysmal abdominal pain and migraine-like symptoms in patients. To facilitate this complex clinical diagnosis, physicians should include an EEG with 24 hr monitoring.

Broader Clinical Implications

The lengthy period during which the patient in the case study suffered needlessly prior to resolution of her rare condition highlights the need for perseverance in thoroughly assessing seemingly "mysterious" pain conditions as opposed to attributing them to purely psychological factors. Despite the progressive rejection of the mind-body dualism of Freud and others that has profoundly affected our views of rare pain disorders that are difficult to assess, it is only recently that more clearly defined pathophysiological and neurobiological bases of functional pain conditions have become understood. In this case, attribution of psychogenic (i.e. purely due to psychological factors) pain could have resulted in the patient spending the rest of her life without adequate pain relief, inappropriate treatments, iatrogenic harms, and additional stigmatization. Historically, the Freudian psychoanalytic model regarded chronic pain and somatic disorders as thinly disguised psychological issues. The medical field of antiquity traditionally regarded conditions such as migraine, asthma, arthritis, hypertension, diabetes, and tuberculosis as syndromes of exclusively psychogenic etiologies,[5] such as repressed emotions or an anxious disposition. As a classic example, neurologists have now abandoned the notion of "migraine personality types." As modern medicine has moved away from the psychogenic model of these conditions and biological bases have been established, the biopsychosocial model has taken hold. This model is contigent on the interconnectedness of biology, psychology, and socio-environmental factors—all of which influence disease pathology. However, within the biopsychosocial framework, disease is regarded as a biologically-based phenomenon that can both be influenced by psychology and can also cause psychological outcomes,[18] while making no assertion regarding underlying psychological causes.

While many conditions have made the leap from the psychogenic to the biopsychosocial model, other rare conditions, such as abdominal epilepsy or a number of chronic pain conditions are still too often relegated to the status of psychological disorders. While falling into the trap of the psychogenic model may assuage the practitioner unable to explain the etiology of a patient's symptoms, this comfort may come at the expense of the patient's quality of life, dignity, and future wellbeing. Pickoff[19] recently warned of the perils of "psychological mislabeling," referring to the faulty assumption that depression, anxiety, and stress are routinely the cause of poorly understood chronic conditions including painful ones. Although the biopsychosocial model can have its benefits, he notes that it is most likely evoked when faced with our most challenging patients. By failing to take the myriad physical symptoms seriously from the onset, the precious window of time during which interventions can abate disease progression quickly dwindles. The patient in question was not able to successfully mitigate her abdominal epilepsy and concomitant lethargy until the age of fifty-two, prior to which she was compelled to treat her pain with codeine/paracetamol on a prn basis. Finally, at the age of fifty-two, her symptoms have been largely alleviated with carbamazepine and, seemingly more critical, a therapeutic dosage of pregabalin.

As illustrated by the case in question, a patient presenting with medically unexplained symptoms, even those which seem to be bidirectionally modulated by psychological factors, does not justify a psychological evaluation as the sole avenue for treatment. This approach inherently places the blame on the patient for his or her symptoms.[20] In regard to chronic pain conditions or rare disease presentations such as abdominal epilepsy, physicans must be willing to acknlowdge their own educational and clinical shortcomings. The patient's phenomenological experience of pain, whatever the cause, deserves the astute consideration and thoughtful professional care of his or her treatment team.

Disclosure

The authors report no conflicts of interest in this work.

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