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Nocturnal generalized seizures preceded by a gastrointestinal aura: a case report

Despite the fact that abdominal pain is a common symptom of various pathological conditions, it can also be suggestive of a diagnosis of epilepsy. Autonomic epilepsy may present with paroxysmal gastrointestinal symptoms mimicking abdominal pathology or functional disorders, making diagnosis challenging. In 36 cases reported in the last 34 years, the most common gastrointestinal symptoms included abdominal pain, nausea, vomiting, etc. Although many mechanisms have been outlined, the cause of autonomic epilepsy with gastrointestinal symptoms still remains unclear current hypotheses suggest that the Sylvian fissure and the insular cortex may serve as sources of epileptic activity. Recognition of autonomic epilepsy in such casesis important, as timely identification and antiseizure treatment lead to significant clinical improvement.

Case presentation. A female patient presented with recurrent nocturnal episodes beginning with abrupt awakening due to painful urges to defecate, followed by loss of consciousness, generalized tonic–clonic seizures and postictal confusion.

Initially rare, the episodes increased to 4—5 events per year, exclusively during sleep,with normal neurological status between seizures. Brain MRI revealed bilateral cortical atrophy of the frontal and temporal lobes, asymmetry of the lateral ventricles, and periventricular leukoaraiosis. Overnight videoEEG revealed interictal sharp waves during sleep without recorded ictal events. Levetiracetam therapy (500 → 750 mg/day) led to complete seizure control.

Discussion. In this case, the combination of nocturnal onset, with gastrointestinal manifestations, and subsequent generalized tonic—clonic activity suggests either a focal seizure manifesting as gastrointestinal pain with secondary generalization or a generalized seizure preceded by a gastrointestinal aura. VideoEEG monitoring confirmed the epileptogenic nature and contributed to understanding the pathophysiology of these symptoms. Levetiracetam proved effective in achieving seizure freedom.

Conclusions. This case highlights a rare epileptic presentation with nocturnal seizures preceded by a painful gastrointestinal urge. Recognizing such visceral symptoms may aid earlier diagnosis and appropriate treatment.

Keywords: autonomic epilepsy, visceral aura, gastrointestinal symptoms.

Despite the fact that abdominal pain is a common symptom of various pathological conditions, it can also be suggestive for a diagnosis of epilepsy. Autonomic epilepsy is an uncommon syndrome in which paroxysmal gastrointestinal symptoms can result from seizure activity. It is characterized by otherwise unexplained, paroxysmal gastrointestinal complaints, symptoms of a central nervous system disturbance, an abnormal electroencephalography (EEG) with findings specific for a seizure disorder, and improvement with antiseizure medication. In 36 cases reported in the last 34 years the most common gastrointestinal symptoms include paroxysmal abdominal pain, nausea, and vomiting, sometimes accompanied by bloating and diarrhoea, while the most common neurological symptoms include lethargy and confusion, more rarely — headache, syncope and transient blindness [3, 6, 7, 11]. As a type of autonomic epilepsy, such condition can also be associated with some autonomic phenomena co-relating with the episodes such

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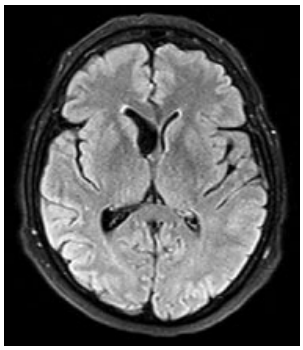

Fig. 1. *Brain MRI, FLAIR sequence, axial projection*

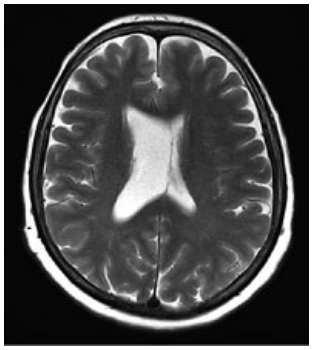

Fig. 2. *Brain MRI, T2-weighted sequence, axial projection*

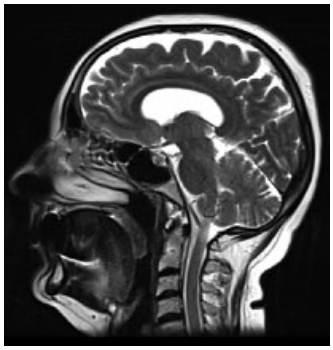

Fig. 3. *Brain MRI, T2-weighted sequence, sagittal projection*

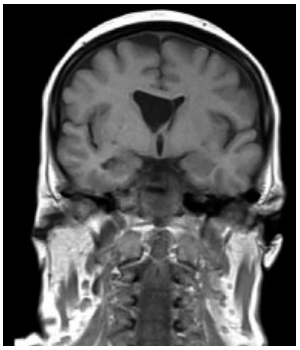

Fig. 4. *Brain MRI, T1-weighted sequence, coronal projection*

as pallor or cold sweating [10]. Abdominal auras can also be seen with manual and oral automatisms, that is, an auto motor seizure [4].

It is known that epileptic auras represent the initial subjective component of a seizure and provide valuable information about the localization of the epileptogenic zone. Visceral or gastrointestinal auras, although uncommon, are often linked to temporal or insular lobe epilepsy [2].

Although many mechanisms have been outlined, the cause of autonomic epilepsy with gastrointestinal symptoms still remains unclear. There are hypotheses that the Sylvian fissure and insular cortex lying right beneath it could be the origin of the seizure as they coincide with the locations of the abdomen on the Sensory homunculus. Additionally, the M2 portion of the middle cerebral artery courses through the Sylvian fissure. Recurrent abdominal pain may also be seen in visceral hyperalgesia, peptic ulcer disease and abdominal migraine. The most common differential diagnosis for autonomic epilepsy with abdominal aura is abdominal migraine as they have many overlapping features. Duration of the symptoms may be used to differentiate the two entities; the duration is longer in abdominal migraine than in autonomic epilepsy. The EEG is usually abnormal in autonomic epilepsy and may be used to confirm the diagnosis [1].

It is already known that patients with autonomic epilepsy may have normal EEG patterns in the interictal periods, extra-temporal origins of epileptic foci and secondary generalisation. There are strong suggestions that EEG conducted after the first 24 hours after the epileptic episode can detect abnormalities to a greater extent [1].

We report a case of a woman with nocturnal episodes of generalized tonic-clonic seizures consistently preceded by a painful defecatory urge — an unusual presentation that can broaden the differential diagnosis of epilepsy.

Clinical case

A female patient presented with a several-year history of recurrent nocturnal episodes. Each episode began with abrupt awakening from sleep due to intense, painful urges to defecate. Within minutes, she lost consciousness and developed generalized tonic — clonic convulsions, followed by postictal confusion and fatigue lasting several minutes. Initially, these events occurred once every one to two years and were considered as a gastrointestinal pathology. The patient was evaluated by the general practitioner and gastroenterologist, esophagogastroduodenoscopy, colonoscopy, a computed tomography scan for cancer screening and laboratory tests were performed, with no abnormalities found. In 2024, the frequency of these episodes increased to four or five episodes per year. All events took place during sleep; none were reported during wakefulness. There were no preceding triggers such as alcohol intake, stress, or sleep deprivation. Neurological examination between episodes was normal.

Brain magnetic resonance imaging (MRI) revealed cortical atrophy in the frontal and temporal lobes, asymmetry of the lateral ventricles, and periventricular leukoaraiosis (Fig. 1—4).

Overnight video-EEG monitoring was performed. No ictal events were recorded during the monitoring period. Interictal epileptiform discharges were noted during sleep, predominantly in the form of sharp waves (Fig. 5).

Treatment with levetiracetam 500 mg daily was started, resulting in a single breakthrough seizure. After increasing the dose to 750 mg daily, the patient has remained seizure-free to date.

Discussion

Visceral auras involving painful abdominal or rectal sensations are rare and may originate from the insular or mesial temporal regions. Gastrointestinal complaints, in particular abdominal pain, may be the only manifestation of epileptic activity.

In 2013 C. Cerminara et al. described a case of an 8-year-old boy with no family history of epilepsy, who suffered from recurrent episodes of abdominal pain since about 6 months of age. He described the pain as «a sword-piercing», localized mainly in the epigas-

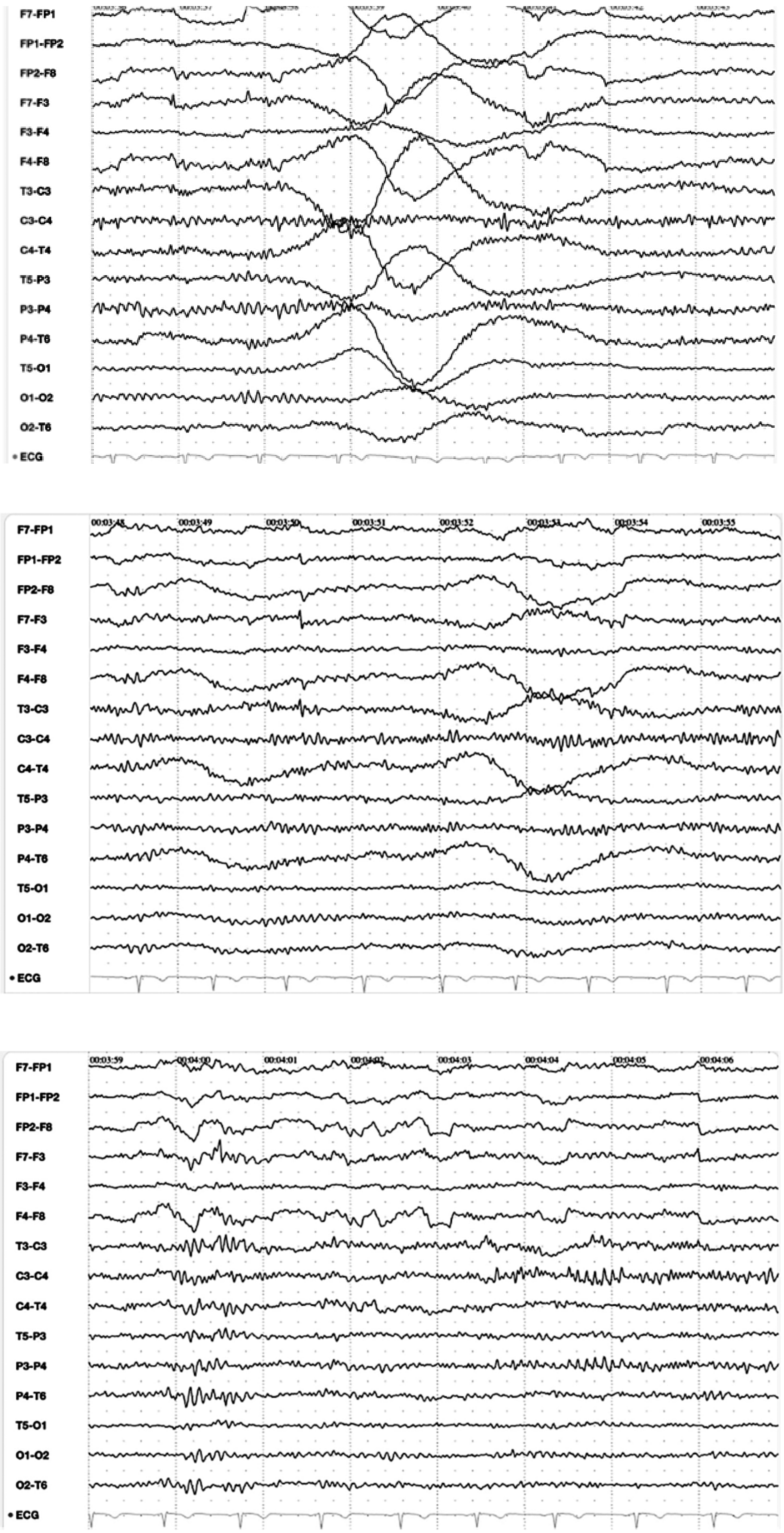

Fig. 5. *Fragments of video-EEG monitoring recorded during drowsiness, subtle focal slowing and intermittent sharp transients over the left centro-temporal region*

tric region and varied from a few minutes to 1 hour, with a frequency of 5—8 episodes per day, almost always associated with pallor and nausea, with sudden onset and spontaneous resolution. There was no impairment of consciousness and he never had convulsions; the paroxysms were followed by increased sleep. He underwent a negative abdominal investigation; neurological examinations and brain MRI were normal. Interictal EEG during wakefulness and sleep displayed bilateral spikes and diphasic sharp-waves over the temporal leads with a marked increase in frequency during drowsiness; a 24-hour EEG-monitoring showed several bilateral synchronous and asynchronous temporal spikes during wakefulness and nocturnal sleep. The ictal EEG during severe abdominal pain in the epigastric region with nausea and pallor showed rhythmic spikes on the centro-temporal regions. The patient started treatment with Carbamazepine (20 mg/kg/day) with a progressive decrease in seizure frequency. At the last follow-up, when he was 9-years old, he was seizure free [2].

A. Balabhadra, et al. also described a case of a 20-year-old man with recurrent 8—12 minute episodes of severe «piercing» epigastric abdominal pain 3—5 times per day for six months associated with non-projectile vomiting followed by lethargy or loss of consciousness which lasted for a «few minutes» after each episode. General and local examinations of the abdomen and nervous system did not reveal any significant findings. He was prescribed analgesics and proton pump inhibitors without any improvement in his symptoms. Brain MRI was normal. Psychiatric examinations didn't reveal any pathology. A 30-minute awake EEG revealed repetitive sharp waves in the right temporal leads which were highly suggestive of an epileptogenic focus arising from the right temporal lobe. He was started on oxcarbazepine 450 mg twice daily, showed a progressive decrease in seizure frequency over a 12-month period and has been asymptomatic throughout the next 4-month period of follow-up [1].

T.G. Phan, et al. reported an unusual case of ictal abdominal pain occurring in the setting of parietal lobe haemorrhage and suggested a possible role of the somatosensory area in pain perception [8]. A supplementary motor area was considered as another possible location for abdominal pain. Occasionally focal epilepsy with ictal abdominal pain has been related to brain tumors and brain disorders [9]. Previous reports on ictal abdominal pain have shown right parieto-occipital encephalomalacia, biparietal atrophy and bilateral perisylvian polymicrogyria [5].

The nocturnal occurrence of such episodes can complicate differentiation from sleep disorders, syncope, or parasomnias. The main differential diagnosis of autonomic epilepsy presenting with abdominal aura is abdominal migraine, as the two conditions share several overlapping features. EEG and episode duration may help distinguish between them, with abdominal migraine typically lasting longer than epileptic events. Other important differentials include irritable bowel syndrome, vasovagal syncope and symptomatic (provoked) seizures. As well, in contrast to functional gastrointestinal disorders and reflex syncope, autonomic epileptic seizures are usually associated with epileptiform abnormalities on EEG, which may support the diagnosis. Recognition of this presentation is crucial, as patients may initially present to gastroenterologists or general practitioners due to the misleading visceral symptoms. Unexplained paroxysmal gastrointestinal complaints, impairment of consciousness, and focal abnormal EEG are the main criteria to establish a diagnosis of focal epilepsy with ictal abdominal pain, but not all the criteria need to be present in each case. The abrupt onset, the spontaneous resolution, and the relatively short duration of episodes may be helpful for a correct and early diagnosis. We must also be aware of the stigma that revolves around the diagnosis of a somatoform disorder in these scenarios and quickly consider getting an EEG, preferably a video-EEG if the facilities are available for such patients so that their symptoms are not falsely labelled as «psychogenic» or «functional». If the EEG findings are abnormal, the treatment involves the initiation of antiepileptic drugs with regular follow-up [1]. In this case, the combination of nocturnal onset, gastrointestinal symptoms, and subsequent generalized tonic—clonic activity suggests either a focal seizure manifesting as gastrointestinal pain with secondary generalization or generalized seizure preceded by a gastrointestinal aura. Video-EEG monitoring confirmed the epileptogenic nature and contributed to understanding the pathophysiology of these symptoms. Levetiracetam proved effective in achieving seizure freedom, consistent with current recommendations for focal and generalized epilepsies.

Conclusions

This case illustrates an uncommon epileptic manifestation characterized by nocturnal seizures preceded by a painful gastrointestinal urge. Awareness of such visceral symptoms can improve diagnostic accuracy and timely initiation of appropriate antiseizure therapy.

There is no conflict of interest.

Authors' participation: data collection, data analysis and interpretation, results processing — D.I. Hnatovska, O.P. Lebed, G.V. Perkova; writing the original draft — D.I. Hnatovska; data collection, conceptualization, methodology, results processing— Yu.O. Solodovnikova; conceptualization, methodology, results processing — V.V. Dobrovolskyj, A.S. Son.

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Автономна епілепсія з гастроінтестинальною аурою та нічними генералізованими нападами: клінічний випадок

Незважаючи на те, що абдомінальний біль є поширеним симптомом різноманітних патологічних станів, у окремих випадках він може бути проявом епілепсії. Автономна епілепсія може проявлятися пароксизмальними гастроінтестинальними симптомами, що імітують абдомінальну патологію або функціональні розлади, ускладнюючи діагностику. У 36 випадках, описаних упродовж останніх 34 років, найчастішими гастроінтестинальними симптомами були абдомінальний біль, нудота, блювання тощо. Незважаючи на запропоновані патофізіологічні механізми, етіологія автономної епілепсії з гастроінтестинальними проявами залишається остаточно не з'ясованою; згідно з наявними гіпотезами, можливими джерелами епілептичної активності є ділянка сильвієвої щілини та острівцева кора. Раннє розпізнавання таких станів є критично важливим, оскільки своєчасне призначення протинападової терапії є критично важливим для покращення стану пацієнта.

Клінічний випадок. Пацієнтка звернулась зі скаргами на повторювані нічні епізоди протягом кількох років, що починались з раптового пробудження через болісні позиви до дефекації, з подальшою втратою свідомості, розвитком генералізованого тоніко-клонічного нападу та постіктальної сплутаності свідомості. Частота епізодів з часом зросла до 4—5 на рік, вони виникали виключно під час сну, змін у неврологічному статусі між нападами не спостерігалось. Магнітно-резонансна томографія головного мозку виявила білатеральну кортикальну атрофію лобних та скроневих часток, асиметрію бічних шлуночків та перивентрикулярний лейкоареоз. Під час нічного відео-ЕЕГ-моніторингу виявлено інтеріктальні гострі хвилі у фазі сну без реєстрації іктальних подій. На тлі терапії леветирацетамом (500 → 750 мг/добу) досягнуто повного контролю нападів.

Обговорення. Поєднання нічного дебюту, гастроінтестинальних симптомів і наступних генералізованих тоніко-клонічних нападів у цьому випадку свідчить на користь фокального нападу з гастроінтестинальними проявами та вторинною генералізацією або генералізованого нападу з гастроінтестинальною аурою. Відео-ЕЕГ-моніторинг підтвердив епілептогенну природу епізодів і дозволив уточнити їхні патофізіологічні механізми. Призначення леветирацетаму забезпечило повний контроль нападів.

Висновки. Цей випадок ілюструє рідкісний варіант епілепсії з нічними нападами, яким передують гастроінтестинальні симптоми. Розпізнавання подібних вісцеральних симптомів може сприяти ранній діагностиці та своєчасному призначенню адекватної терапії.

Keywords: автономна епілепсія, вісцеральна аура, гастроінтестинальні симптоми.

ДЛЯ ЦИТУВАННЯ

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