Focal epilepsy with ictal abdominal pain: a case report

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Abstract

Focal epilepsy with ictal abdominal pain is an unusual partial epilepsy characterized by paroxysmal episodes of abdominal or visceral pain, disturbance of awareness and electroencephalographic abnormalities. We describe a new case of ictal abdominal pain in which gastrointestinal complaints were the only manifestation of seizures and review the previously described pediatric patients. In our patient clinical findings, ictal EEG abnormalities, and a good response to antiepileptic drugs allowed us to make a diagnosis of focal epilepsy with ictal abdominal pain. This is a rare epileptic phenomenon that should be suspected in patients with unexplained paroxysmal abdominal pain and migraine-like symptoms. We suggest that, after the exclusion of more common etiologies, focal epilepsy with ictal abdominal pain should be considered in patients with paroxysmal abdominal pain and ictal EEG abnormalities.

Introduction

Recurrent episodes of abdominal pain are common in children and adults. Several pathological conditions can lead to paroxysmal gastrointestinal symptoms, such as porphyria, cyclical vomiting, intestinal malrotation, peritoneal bands, and abdominal migraine [1]. Psychological and emotional factors may also play an important role in some patients with gastrointestinal disorders. However, in a number of patients the episodic nature of abdominal pain can be suggestive for a diagnosis of epilepsy [1]. Epileptiform EEG abnormalities, loss or alteration of consciousness, and a good response to antiepileptic drugs are other features that can lead to a diagnosis of focal epilepsy with ictal abdominal pain [2,3]. We describe one child affected by epilepsy which had recurrent and severe abdominal pain as the only manifestation of epileptic seizures.

Case report

An 8-year-old boy was born at 39 weeks of gestation by selective cesarean section. The pregnancy was complicated by a sudden reduction in fetal heart rate. All developmental milestones were regularly achieved. There was no family history of epilepsy. The boy experienced recurrent episodes of abdominal pain since about 6 months of age. He described the pain as “a sword that pierces my belly”, localized mainly in the epigastric region and its duration varied from a few minutes (more frequently) to 1 hour, with a frequency of 5–8 episodes per day. The intense abdominal pain was almost always associated with pallor and nausea, but not accompanied by scream or cry. The attacks were sudden in onset and had spontaneous resolution. There was no impairment of consciousness, also in longer episodes, and he never had convulsions; the paroxysms were followed by increased sleep. He underwent a negative abdominal investigation including complete blood count, stool examinations for ova and parasites, abdominal ultrasound and upper gastrointestinal endoscopy. Physical and neurological examinations were normal.

Intercital EEG during wakefulness and sleep displayed bilateral spikes and diphasic sharp-waves localized over the temporal leads with a marked increase in frequency during drowsiness. A 24-hours EEG recording showed several bilateral synchronous and asynchronous temporal spikes during wakefulness and nocturnal sleep. At 9:30 in the morning a seizure characterized by severe abdominal pain in the epigastric region with nausea and pallor was recorded. The ictal EEG showed rhythmic spikes on the centro-temporal regions (Figure 1). Magnetic resonance imaging of the brain was normal. The patient started treatment with Carbamazepine (CBZ) (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.

**Discussion**

Epigastric sensations are frequent symptoms in patients with partial epilepsy and may include abdominal pain, nausea, vomiting and hunger, and have been reported to be the most common aura in temporal lobe epilepsy [3-5]. Painful epileptic auras were reported in 4.1% of 25 patients with focal epilepsy by Nair et al. [6]. Abdominal pain was present in 5% of all abdominal auras in temporal lobe epilepsy and 50% in frontal lobe epilepsy [6]. However, gastrointestinal complaints, in particular abdominal pain, may be the only manifestation of epileptic activity [1,3,4,7]. 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 [2,3,6]. In addition, a variety of migraine-like disturbances such as nausea, headache, dizziness, and visual hallucinations may be associated with pain during the attacks [4]. When the migraine-like symptoms are present it is often difficult to differentiate focal epilepsy with ictal abdominal pain from migraine or other neurological disorders, such as Panayiotopoulos syndrome. The abrupt onset, the spontaneous resolution, and the relatively short duration of episodes may be helpful for a correct and early diagnosis of focal epilepsy with ictal abdominal pain. Another helpful distinguishing feature of epilepsy with severe abdominal pain could be the localization of ictal pain, that is most commonly periumbilical or upper abdominal and rarely spreads to involve other body parts, such as in our patient [1-4]. EEG abnormalities have been reported in most patients with focal epilepsy and ictal abdominal pain [1,4]. Few reports described ictal EEGs: during the seizure the EEG often shows a runs of high voltage slow waves and generalized spike and wave discharges [2-4,8]. In our patient, 24-hours EEG was suggestive of a focal onset, as in two reports that showed clear focal EEG changes over the left hemisphere during an episode of abdominal pain [9,10]. Table 1 shows the clinical characteristics of our patient and the previous pediatric cases described in literature (Table 1) [1-5,7-13].

The pathophysiology of focal epilepsy with ictal abdominal pain remains unknown. Abdominal sensations reproduced by stimulating the insula and sylvian fissure, suggest that these areas may have an important role in explaining the origin of focal epilepsy with ictal abdominal pain [3]. Phan et al. [14], 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. 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 [2,8]. Previous reports on ictal abdominal pain have shown right parieto-occipital encephalomalacia, biparietal atrophy and bilateral perisylvian polymicrogyria [9].

In conclusion, our patient showed recurrent attacks of severe abdominal pain as the only manifestation of epileptic seizure. Focal epilepsy with ictal abdominal pain is a rare epileptic phenomenon that should be suspected in patients with unexplained paroxysmal abdominal pain and migraine-like symptoms. The correct diagnosis at the onset may be difficult to establish; in these cases prolonged EEG recordings with 24-hours monitoring must be considered to facilitate the clinical diagnosis.
| Patient number | Age | Sex | Gastrointestinal symptoms | Other non-gastrointestinal symptoms | Episode duration | EEG | Treatment | Outcome |
|----------------|-----|-----|---------------------------|------------------------------------|------------------|-----|-----------|---------|
| Zdraveska N. et al.¹ | 14 | F | Colicky epigastric pain, nausea, vomiting and diarrhea | Pallor, dizziness | 10-30 minutes | Spikes, sharp waves over the right central and temporal regions with secondary generalization | Carbamazepine | Seizure free |
| Franzon RC et al.² | 6 | F | Abdominal pain | Disturbed awareness, occasional generalized tonic-clonic seizures | Seconds to minutes | Spikes and slow waves over left temporal area | Anticonvulsants, surgical resection of oligoastrocytoma | Seizure free |
| Garcia-Herrero D. et al.³ | 14 | F | Colicky periumbilical pain | Headache, pallor, dizziness, multicolored photopsia | Second to minutes | Interictal-bursts of sharp and slow waves | Valproic acid | Near complete resolution |
| Dutta SR et al.⁴ | 15 | M | Epigastric abdominal pain and vomiting | Lethargy | 30 minute to hours | Right temporal focal seizure discharge with generalization | Oxcarbazepine | Seizure free |
| Case 1 | | | | | | | |
| Dutta SR et al.⁴ | 13 | F | Colicky periumbilical pain | NR | 10-30 minutes | Generalized spikes and wave discharges | Oxcarbazepine | Seizure free |
| Case 2 | | | | | | | |
| Young GB et al.⁵ | 15 | F | Abdominal pain | Generalized tonic seizures | NR | Multiple independent spikes | NR | NR |
| Hasan N. et al.⁷ | 8 | M | Colicky periumbilical pain, vomiting | Pallor, an episode with jerky movements in the lower limbs | 10-30 minutes | Generalized paroxysmal epileptiform activity, maximum on photic stimulation | Valproic acid | Seizure free |
| Siegel AM et al.⁸ | 1 | F | Crampy periumbilical pain | Occasional generalized seizures | Few seconds | Right parietal focus | NR | NR |
| Mitchell WG et al.⁹ | 6 | M | Vomiting | Bad smell, fatigue | 20-40 seconds | Ictal and interictic high voltage arrhythmic delta waves, sometimes sharply contoured | Multiple antiseizure medication, than surgery and radiation (for astrocytoma) | Decreased frequency of episodes |
| Douglas EF et al.¹⁰ | 11 | F | Paroxysmal, periumbilical abdominal pain | Lassitude, post-ictal sleep, fever, headache, confusion. “Brief” | | Irregular 3 Hz spike-waves activity | Phenobarbital | Seizure free |
| Case 1 | | | | | | | |
| Douglas EF et al.¹⁰ | 5 | F | Crampy, paroxysmal abdominal pain | Lethargy, post-ictal sleep | Few minutes | Episode 6-7 activity in L temporal area, burst of generalized irregularly intermixed spikes and slow waves | Phenobarbital | Lost to follow-up |
| Case 2 | | | | | | | |
| Douglas EF et al.¹⁰ | 6 | M | Paroxysmal pain | Lethargy, confusion, fever | Few minutes | Paroxysmal spike-wave activity, frontal or generalized | Anticonvulsivants | Seizure free |
| Case 3 | | | | | | | |
| Yingkun F¹¹ | 3 | M | Abdominal pain, vomiting | Confusion, cyanosis, urinary incontinence, blindness | Few minutes | Scattered high voltage slow activity and high voltage sharp waves | Phenytoin, phenobarbital | Seizure free |
| Case 1 | Yingkun F et al.11 | 16 M | Upper abdominal pain, nausea | Disturbance of consciousness | 3-5 minutes | High voltage slow waves; high voltage sharp waves with hyperventilation | Phenobarbital | Seizure free |
| Case 2 | Yingkun F et al.11 | 11 F | Periumbilical abdominal pain | Disturbance/loss of consciousness | Minutes to hour | Bilateral high voltage spikes, complexed slow waves | Phenytoin, phenobarbital | Seizure free |
| Case 3 | Singhi PD et al.12 | 10 M | Periumbilical pain | Pallor, sweats, lethargy, post-ictal sleep | Few minutes | Sharp spikes, spikes and wave activity arising over the central region and becoming generalized | Phenytoin | Complete resolution |
| Agrawal P. et al.13 | 6 M | Colicky periumbilical pain | Lassitude, post-ictal sleep | Half an hour | Generalized slowing, right posterior spikes | Carbamazepine | Seizure free |
| Our patient | 8 M | Colicky epigastric pain, nausea | Pallor | Few minutes to 1 hour | Bilateral synchronous and asynchronous spikes and diphasic sharp-waves in temporal and central area, increased during drowsiness and sleep | Carbamazepine | Seizure free |
Consent
Written informed consent was obtained from the patient’s parents for the publication of this report.

Competing interests
The authors declare that they have no financial and non-financial competing interests.

Authors’ contributions
CC (Medical Doctor) drew the first draft with the assistance and contribution of NEM (Medical Doctor); DR (Medical Doctor) reviewed relevant articles on the literature under the supervision of PC (Director of the Department of Pediatric Neuroscience Unit); PC revised the final draft. All authors contributed to the intellectual contents and approved the final version.

Received: 7 October 2013 Accepted: 4 December 2013
Published: 9 December 2013

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doi:10.1186/1824-7288-39-76
Cite this article as: Cerminara et al.: Focal epilepsy with ictal abdominal pain: a case report. Italian Journal of Pediatrics 2013, 39:76.