

## Case Report

## Epileptic Angina☆

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## ABSTRACT

**Purpose:** To investigate the probable ictal origin of unexplained episodic chest pain and if possible to lateralize and localize the epileptic focus.

**Methods:** A 14 year old boy presented with episodic short lasting localized chest pain. His cardiac and other systemic work-up were normal. MRI brain did not reveal any structural pathology. Video telemetry was done for characterization of the paroxysms.

**Results:** Interictal record showed left fronto-central epileptiform discharges. A left hemispheric, predominantly centroparietal ictal rhythm was identified. The possible localizations of this unusual semiology are somatosensory areas I and II, supplementary sensorimotor area, posterior insula and cingulate cortex. Patient responded remarkably to antiseizure drugs.

**Conclusion:** Pain is a rare manifestation of epilepsy observed in less than 1% of patients. When present, it is usually accompanied by other focal features. This rare occurrence of epileptic seizures masquerading as angina is a novel observation.

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## 1. Introduction

Among the manifold unusual semiologies of epilepsy, pure epileptic chest pain has not received attention to date. Pain occurs in a small minority of patients with epilepsy but is often accompanied by other motor, sensory or behavioural features which denote their ictal origin. The occurrence of epileptic pain in isolation can be a challenging clinical situation. The clinical history and video-EEG features of a young boy, who presented with episodic chest pain, is described to illustrate this atypical presentation.

## 2. Case report

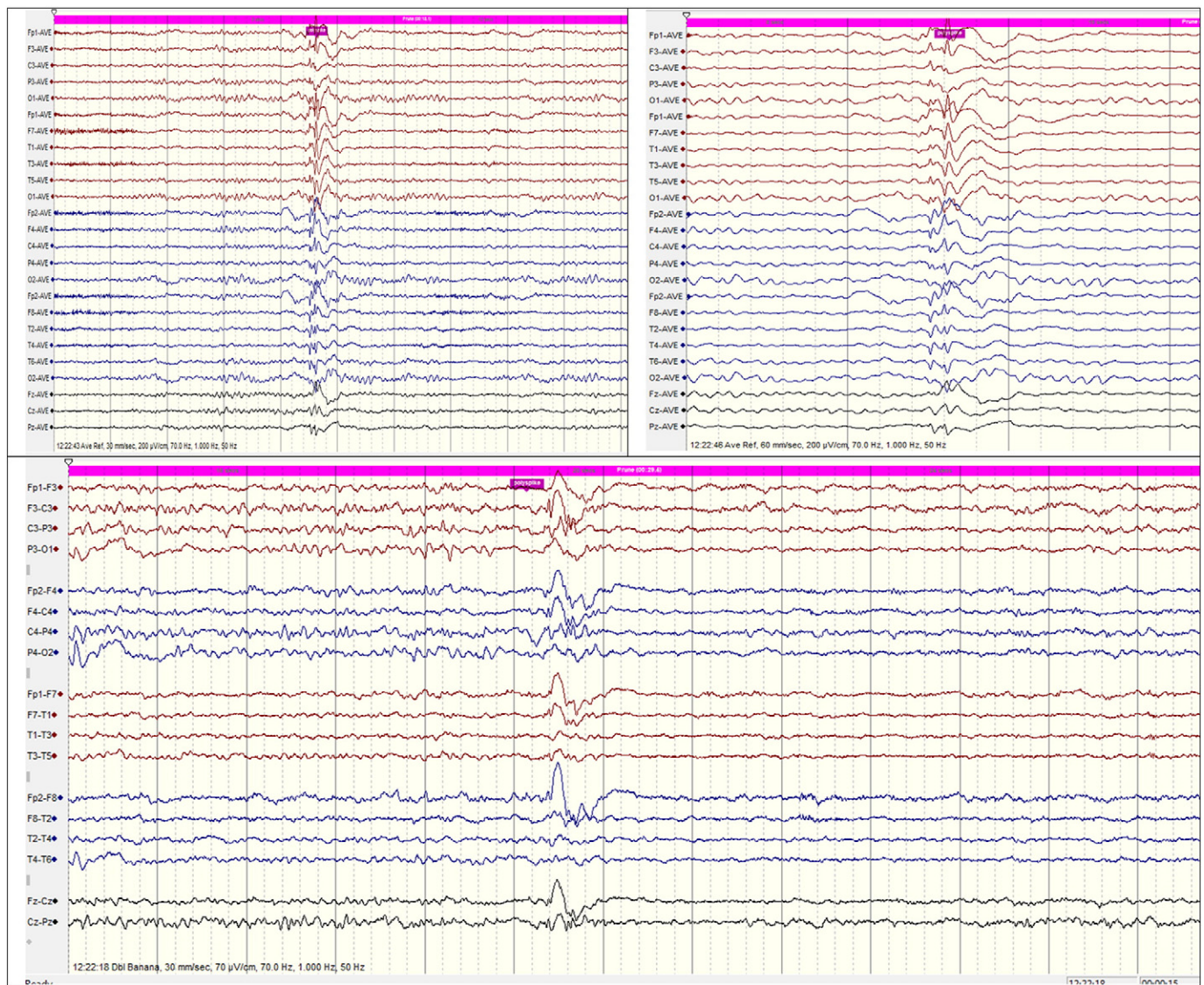
A 14-year-old boy preparing for his final exams was brought by his mother for evaluation of episodic left-sided chest pain of 2 weeks duration. The pain which lasted a few seconds to half a minute was described as sharp, excruciating and localized to the lower chest on the left side and involved an area of only two finger

breadths. The frequency varied from 1 to 8 episodes per day with variable severity but consistent location. There were no accompanying clinical features like breathlessness, diaphoresis, palpitation, fear, abnormal behavior or movements. There was no antecedent illness or trauma; no history of febrile seizures, encephalitis or perinatal insult. His scholastic performance was good and his behaviour with peers and family was normal. There was no family history of seizures or any other major illness. He was evaluated by a pulmonologist and cardiologist who performed relevant investigations including a chest CT scan, electrocardiogram, and echocardiogram which returned normal findings. He was referred to a psychiatrist but the family preferred to get an opinion at our Neurology Center. An interictal EEG was initially performed which showed spike-and-wave discharges in the awake state distributed frontocentrally over the left hemisphere (Fig. 1). This prompted us to obtain a long term video-EEG recording. During the 24 h of study, five events were recorded of which only one was associated with a clear-cut ictal pattern. The clinical semiology was dominated by severe localized chest pain in the right lower chest (as opposed to the typical left-sided location for chest pain). However some paucity of movements were observed on the right side especially the right upper limb. Based on these observations, a left hemispheric focus was postulated. The initial part of the ictal recording was obscured by artifact while a nicely evolving 4.0–4.5 Hz rhythm was noted in the parasagittal region a few seconds after

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**Fig. 1.** Interictal EEG record: Common average referential montage at normal and slower paper speed (upper panel) shows left hemispheric poly spike and wave discharges along with the right hemisphere showing positivity at the onset; the bipolar montage (lower panel) shows more prominent anterior distribution especially frontocentral.

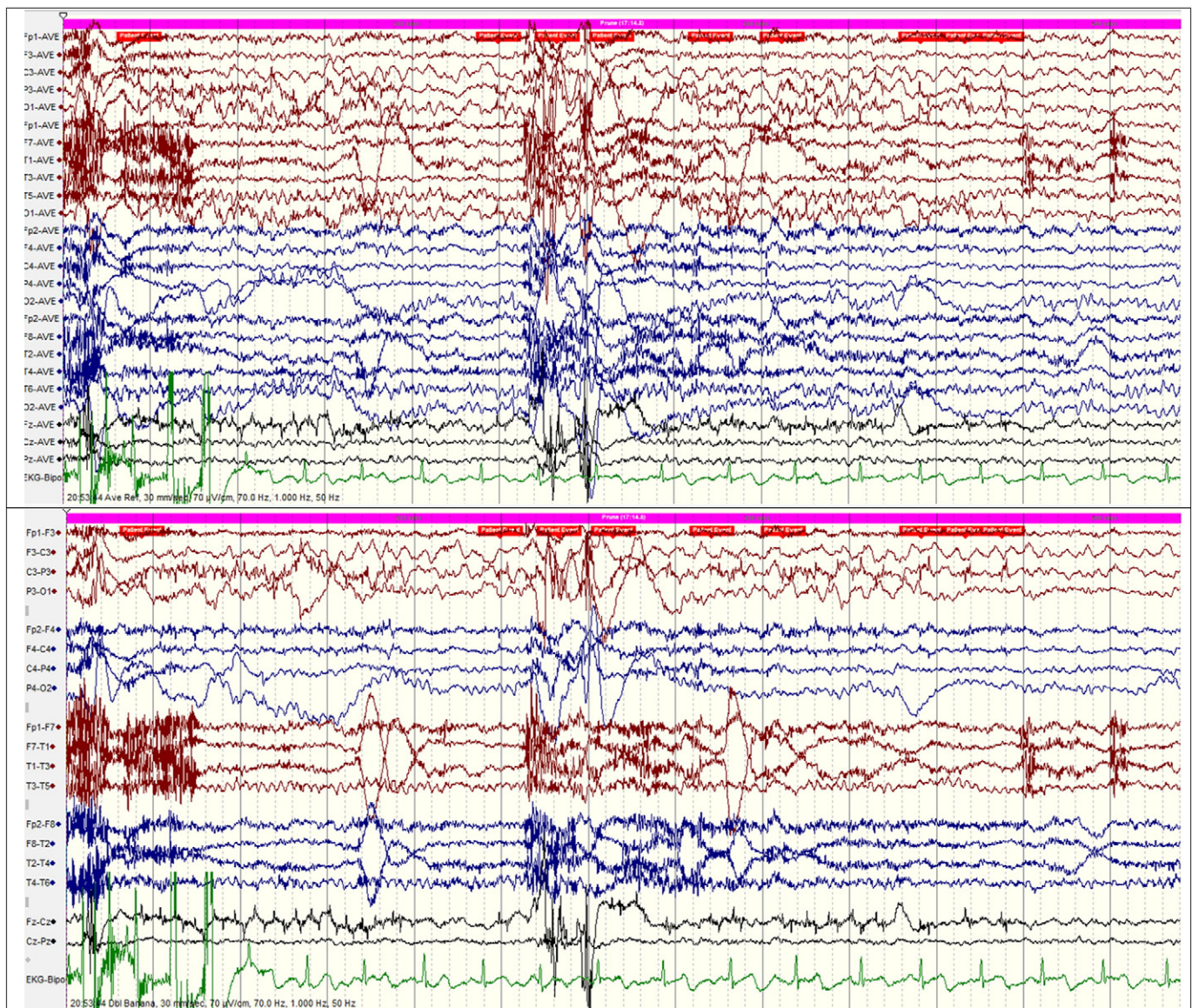
clinical onset. A 9–10 Hz faster rhythm was also observed in the left mid and posterior temporal regions which did not show evolution (Fig. 2). A 1.5 Tesla MRI of the brain did not reveal any focal pathology. The electroclinical data offered definitive proof for the ictal origin of the phenomenology from the left hemisphere. The events completely disappeared after initiation of 100 mg per day of zonisamide. This drug was chosen as first line, because the family wanted seizure freedom in a short time as his exams were due in two days. Later on, he was switched to 400 mg per day of carbamazepine. He has had no seizures, behavioural or cognitive abnormalities during the last four months of follow-up.

### 3. Discussion

Epileptic pain as a somatosensory manifestation of seizures is not uncommon but as a solitary presentation of epilepsy is extremely rare and diagnostically challenging. Pazarci et al. who searched for this elusive symptom in their data base of 4736 patients identified only 9 patients [5]. Their experiences consisted of nuchal pain, headache, abdominal pain and pain in the extremities (peripheral pain). The

ictal EEG more often showed hemispheric or diffuse abnormalities than focal well-defined ictal activity. In our patient the lateralization was clearly evident compared to localization although the parasagittal region showed the most unequivocal changes. All the patients in their study who had lateralized peripheral pain had an abnormal MRI while our report describes MR-negative “algic” seizures. Structural abnormalities were a universal feature of patients with peripheral pain in a previous series of 8 patients with ictal pain. One of the patients had only biparietal atrophy evident on MRI whose SPECT study corroborated the finding. The electrographic abnormalities were either restricted to the parietal area or hemisphere on the surface EEG. Intracranial recordings in three patients showed seizure onset from inferior parietal lobule/parietal operculum or medial parietal lobe [6]. In another analysis of retrospective data of more than five thousand patients, the authors found pain associated with seizures associated with epilepsy in 10 patients of whom 8 had peripheral location of pain. Three of their patients had a normal MRI while two had a central location of ictal onset which is comparable to our observation. However none of these patients in any of the above mentioned series had pain as the sole manifestation of epilepsy thus making our observation a novel one [2]. The major studies which address epileptic pain





**Fig. 2.** Ictal EEG record showed a 4–5 Hz parasagittal rhythm; while a faster 9–10 Hz activity was seen in mid and posterior temporal region in the left hemisphere.

due to seizures associated with epilepsy are summarised in Table 1 [2, 5–7]. None of the patients in these reports had isolated chest pain as an ictal manifestation which makes this case unique and challenging.

The patient had several events during the long term recording without any EEG abnormalities until he had an event with definite ictal abnormalities. This emphasizes the need for a high index of suspicion to discover the ictal origin of unexplained pain. Pain has a localization more or less similar to somatosensory aura. When the seat of origin is the primary sensory area, a “sensory march” may be encountered associated with other elementary motor manifestations. Secondary sensory area (SSA-2) involvement produces vague or complex sensations which are more diffuse in distribution. Posterior insular involvement can also produce sensory phenomena like warmth or dysesthesias which encompasses a widespread cutaneous distribution [3]. Supplementary somatosensory-motor area and cingulate cortex are the other anatomical sites whose involvement can result in sensory manifestations [1]. Pain as such has not been reported in cingulate epilepsy. Given the paracentral distribution, mesial parietal and cingulate localizations need to be considered.

The ictal origin of pain in the present report was established by the presence of electrographic abnormalities and response to antiseizure drugs. This bears resemblance to “ictal epileptic headache” observed especially in the pediatric age group diagnosis of painful seizures which is based on similar grounds [4].

#### 4. Conclusion

Pain as a manifestation of epilepsy is rare. The location and nature of ictal pain is variable as evidenced in this report of pure left-sided chest pain. A dedicated evaluation by video-EEG telemetry can be invaluable in establishing the diagnosis in such cases.

#### Appendix A. Supplementary data

Supplementary data to this article can be found online at <http://dx.doi.org/10.1016/j.ebcr.2017.02.001>.

**Table 1**  
“Pain and related ictal manifestations: demographic, clinical, electrographic and radiological correlation”.

	Patients gender	Age of onset	Seizure characteristics	EEG	Neurological examination	MRI brain
Pazarıcı et al. [5]	9 (5 M/4 F)	6 months–52 years	Pain of extremities associated with elementary motor manifestations (3) Abdominal pain (2) Headache (2) Pharyngeal pain, vomiting associated with hemiconvulsion (1)	Frontotemporal/temperoparietal or diffuse hemispheric slowing/sharp waves	Normal (4) Hemi-hypertrophy (1) Homonymous hemianopia and hemiparesis (1) Left leg paresis (1) Hyperactive reflexes (1)	Normal (3) Gliosis (1) Diffuse white matter abnormalities (1) Subependymal heterotopia (1) Perivascular space dilatation (1) Old hemorrhage (1) Bilateral hippocampal atrophy (1) Lesional zone was fronto-central in 11 (15%), parietal in 22 (30%), temporal in 10 (13%), multilobar, vertex in 4 (5%) operculum in 4 (5%)
Tuxhorn [7]	75 (47 M/28F)	Infancy to 45 years	Numbness, tingling, paresthesias, pain associated with elementary motor manifestations Only 2 pts. had pain Head pain (3) Arm or leg pain (7)	Seizure onset zone was fronto-central in 17 (22%), parietal in 24 (32%), temporal in 10 (13%), non-localized in 15(20%), vertex in 5 (6%) and operculum in 4 (5%)	Normal—53% Mild sensorimotor deficits in 21 patients And hemiparesis in 4 patients	
Asadi-Pooya et al. [2]	10(2 M/8 F)	5–60 years		Interictal Normal in 4 Temporal sharp waves/slowing – 3; frontal slowing/spikes—2  Ictal EEG-central onset (2)	NA	Normal—3 NA—2 frontoparietal meningioma in 1 Frontal cortical dysplasia—1 Temperoparietal encephalomalacia—1 Microvascular disease—1 Generalized atrophy—1 Temperoparietal encephalomalacia—1 Parieto-occipital encephalomalacia—1 Nonenhancing parietal lesion—1 Parietal infarct/atrophy—3 Normal
Siegel et al. [6]	8 (7 M/1F)	1–42 years	Sensory aura: burning/cold/throbbing/aching/knife like pain/squeezing sensation Distribution: Upper extremity—4 Cranial—3 Abdominal—1 Associated features: vertigo, nausea, aphasia, paresis, dyscognitive, elementary motor	Ictal- parietal—3 Frontoparietal—2 Right hemispheric—1 Normal—2 Intracranial-parietal operculum—2 Medial parietal—1 Parietal—1 Inconsistent—1	Hemiparesia/hemianopia/cortical sensory loss—1 Other data-not complete	

## References

- [1] Alkawadri R, So NK, Van Ness PC, Alexopoulos AV. Cingulate epilepsy: report of 3 electroclinical subtypes with surgical outcomes. *JAMA Neurol* 2013;70(8):995–1002.
- [2] Asadi-Pooya AA, Asadollahi M, Sperling MR. Ictal pain: occurrence, clinical features, and underlying etiologies. *Epilepsy Behav* 2016;61:59–62.
- [3] Foldvary-Schaefer N, Unnwongse K. Localizing and lateralizing features of auras and seizures. *Epilepsy Behav* 2011;20(2):160–6.
- [4] Parisi P, Verrotti A, Costa P, Striano P, Zanusi C, Carrozzi M, et al. Diagnostic criteria currently proposed for “ictal epileptic headache”: perspectives on strengths, weaknesses and pitfalls. *Seizure* 2015;31:56–63.
- [5] Pazarıcı NK, Bebek N, Baykan B, Gürses C, Gökyiğit A. Reappraisal of epileptic pain as a rare symptom of seizures. *Epilepsy Behav* Feb 2016;55:101–7.
- [6] Siegel AM, Williamson PD, Roberts DW, Thadani VM, Darcey TM. Localized pain associated with seizures originating in the parietal lobe. *Epilepsia* Jul 1999;40(7):845–55.
- [7] Tuxhorn IEB. Somatosensory auras in focal epilepsy: a clinical, video EEG and MRI study. *Seizure Eur J Epilepsy* 2005;14(4):262–8.