

Case Report

Epileptic Angina[☆]Sachin Sureshbabu^{a,*}, Dinesh Nayak^b, Sudhir Peter^c, Chindripu Sobhana^c, Gaurav Mittal^a^a Department of Neurology, St Stephen's Hospital, Tis Hazari, New Delhi 110054, India^b Department of Neurology, Fortis Malar Hospital, Adayar Chennai, India^c Department of Pathology, Metropolis Labs, Emakulum, Kerala, India

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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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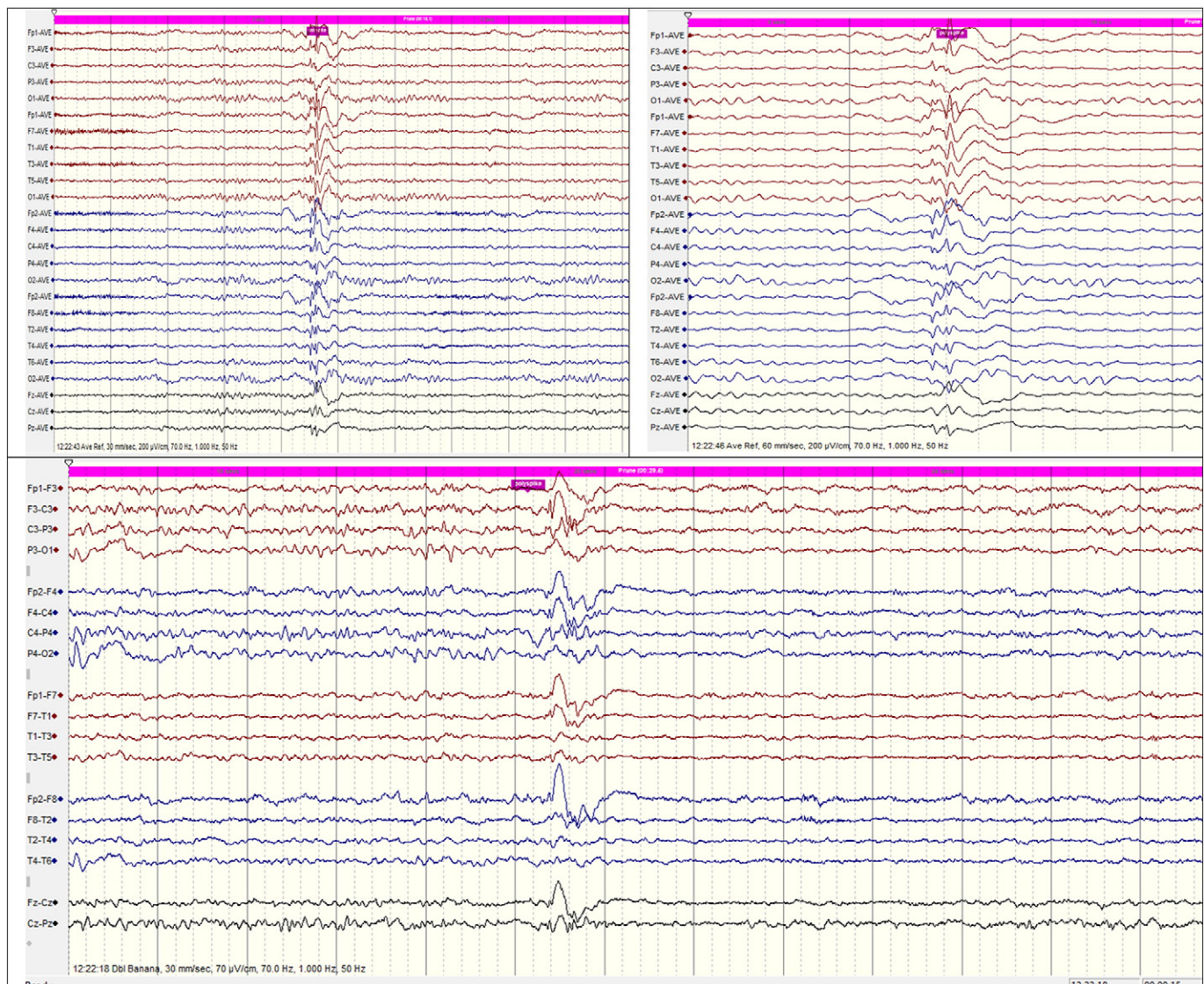


Fig. 1. Intercrictal EEG record: Common average referential montage at normal and slower paper speed (upper panel) shows left hemispheric poly spike and wave discharges along with the right hemisphere showing positivity at the onset; the bipolar montage (lower panel) shows more prominent anterior distribution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

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