CASE REPORT

Reflex gelastic—dacrystic seizures following hypoxic—ischaemic encephalopathy

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SUMMARY

Reflex or stimulus-sensitive epilepsies are uncommon epileptic syndromes triggered by exogenous-specific sensory stimulus or endogenous various mental activities. Gelastic—dacrystic seizures are rare epileptic manifestations characterised by ictal laughter and crying. Gelastic—dacrystic seizures are commonly caused by hypothalamic hamartoma but rarely described due to cortical dysplasia, lesions of frontal and temporal lobes, tumours and vascular malformations. We report a young woman who presented with somatosensory-evoked gelastic—dacrystic seizures. This patient had a positive history of perinatal insult substantiated by MRI findings. Hypoxic—ischaemic encephalopathy as the cause of gelastic—dacrystic seizures has not been reported so far in the literature.

BACKGROUND

Reflex epilepsies are a group of epileptic syndromes in which epileptic seizures are triggered by specific sensory stimuli or certain actions of the patient. Reflex seizures may clinically manifest as partial or generalised seizures. Seizure may be provoked by light, auditory, autonomic, somatosensory, olfactory or other mental activities such as listening music or reading.¹

Gelastic seizures, characterised by bouts of laughter constitute a rare group of epileptic disorders which is commonly associated with hypothalamic hamartoma.² Uncommon causes of gelastic seizures reported in the literature are cortical dysplasia, lesions of temporal and frontal lobes, astrocytomas of mammillary bodies and third ventricular papillomas.³ Dacrystic seizures characterised by ictal crying are also rare epileptic events associated with hypothalamic hamartoma, tumours, vascular malformations, mesial temporal sclerosis or cerebral infarction.⁴

In this case report and videos we present a patient exhibiting tap-induced gelastic-dacrystic seizures associated with hypoxic-ischaemic encephalopathy. Hypoxic-ischaemic encephalopathy as a cause for reflexive gelastic seizures is not described in the literature to the best of our knowledge.

CASE PRESENTATION

A 15-year-old girl presented with a history of convulsions since 5 years. Initially she manifested with abnormal intermittent jerky movements of the left upper limb without loss of consciousness. Her parents noticed falling of objects from the left hand during these episodes. Interestingly these seizures occurred whenever the patient had touched objects. Frequency of these seizures increased with the time. These seizures were triggered by touch or tapping.

Semiology of these seizures could be described as brief tonic-clonic movements accompanied by crying and subsequently bouts of laughter and regaining consciousness within 1–2 min (video 1). These seizures were not provoked by loud voice or light. Perinatal history revealed delayed cry. Milestones were mildly delayed with decreased scholastic performance.

Examination findings disclosed mini-mental status scoring of 27/30 with no focal neurological deficits. Sensory system examination did not reveal any abnormality.

INVESTIGATIONS

Investigations revealed random blood sugar 96 mg/ dL, serum creatine 0.4 mg/dL and haemoglobin 12 g %. Cerebrospinal fluid measles antibody titre was negative. Electroencephalography (EEG) showed periodic lateralised epileptiform discharges over the right hemisphere (figure 1). EEG video of the patient demonstrated gelastic-dacrystic seizure with ictal discharges predominantly in the right parietotemporal areas (video 2). MRI of the brain, T2-weighted and fluid-attenuated inversion recovery (FLAIR) images revealed altered signal intensity suggestive of gliosis and volume loss involving bilateral temporoparietal regions indicating hypoxic-ischaemic encephalopathy (figure 2). On the basis of these peculiar findings somatosensory evoked gelastic-dacrystic seizures was the diagnosis.

TREATMENT

Initial treatment was phenobarbitone (60 mg/day) prescribed by the physician. She poorly responded to therapy. She was then prescribed a combination of sodium valproate (500 mg twice a day) and



Video 1 Video of the patient exhibiting tap induced gelastic dacrystic seizure.

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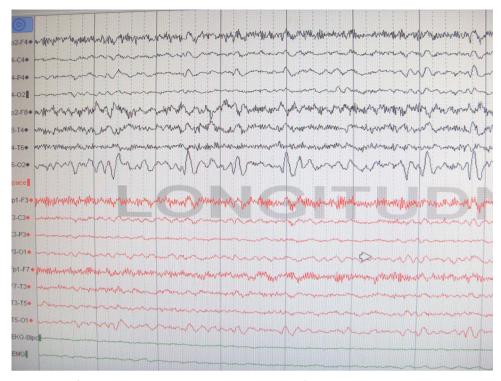


Figure 1 Electroencephalograph of the patient revealing periodic lateralised epileptiform discharges on the right hemisphere.

clonazepam (0.5 mg thrice a day) and once again with poor results. Presently she is on lamotrigine (50 mg twice a day and levetiracetam (500 mg twice a day) resulting in significant reduction of seizures.

OUTCOME AND FOLLOW-UP

At follow-up after 3 months there was more than 50% reduction in seizures.

DISCUSSION

Despite development of the best monitoring technology, hypoxic-ischaemic encephalopathy remains a serious condition that causes significant mortality and long-term morbidity, particularly in developing countries. Hypoxic-ischaemic encephalopathy is one of the most important causes of neonatal seizures. These neonatal seizures usually occur 12–24 h after birth. Approximately 10% of

patients of hypoxic–ischaemic encephalopathy develop epilepsy at the age of 3.5 years. Only some patients develop epilepsy after this age.⁵ In a population-based epidemiological study in children from Brazil, the authors found that symptomatic focal epilepsy was the most common epileptic disorder and hypoxic–ischaemic encephalopathy as its leading aetiological factor.⁶ Our patient presented with reflex seizures after a gap of 10 years of perinatal insult. Reflex epilepsy is an epileptic disorder characterised by provoked seizures due to external stimulus and sometimes due to internal mental mechanism. Various reflex stimuli mentioned in the literature are visual, auditory, olfactory, somatosensory, vestibular, reading, music, internal thoughts and hot water baths.⁷ A case was reported on somatosensory rub-evoked epilepsy originating from the temporal lobe. In this study, the authors described a patient developing seizures evoked by a touch or rub on the



Video 2 Video-EEG demonstrating ictal discharges in right temporal area.

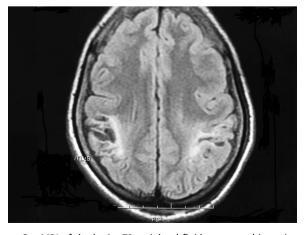


Figure 2 MRI of the brain, T2-weighted fluid-attenuated inversion recovery image showing bilateral gliotic lesions in the parietotemporal areas suggestive of hypoxic–ischaemic encephalopathy.

unilateral arm and shoulder. Single-photon emission CT (SPECT) in the immediate postictal phase disclosed contralateral temporal lobe origin.8 Our patient manifested with gelastic-dacrystic seizures. Gelastic epilepsy is an infrequent seizure disorder described as stereotyped, unprovoked incongruous ictal laughter. The commonest cause of gelastic seizures is hypothalamic hamartoma, although few reports mentioned other aetiologies, like frontal lobe lesions, temporal lobe involvement, parietal lobe disorders and focal cortical dysplasia of cingulate gyrus.³ A patient suffering from gelastic seizures due to involvement of the right parietal lobe has been reported. The seizures described were brief staring followed by smiling and laughing. Seizures originating from the right parietal lobe were substantiated by ictal EEG and SPECT.5 Dacrystic seizures are characterised by ictal crying and rarely associated with gelastic component. Again it is commonly due to hypothalamic hamartoma but rarely dacrystic seizures can occur because of tumours, vascular malformations, hippocampal sclerosis and cerebral infarction. In a recent publication a case with dacrystic seizures due to left temporal lobe involvement was reported. This patient had recurrent complex partial seizures with crying that was reproduced during left carotid methohexital anaesthesia. The author concluded that localised networks involving limbic structures may be involved in patients with dacrystic seizures. 10

Learning points

- ► Gelastic—dacrystic seizures are rare epileptic manifestations characterised by ictal laughter and crying.
- Gelastic-dacrystic seizures are commonly caused by hypothalamic hamartoma.
- Hypoxic-ischaemic encephalopathy can rarely cause gelastic-dacrystic seizures.

Contributors RV made the hypothesis and HNP helped in drafting the final version of the manuscript.

Competing interests None.

Patient consent Obtained.

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