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Case Report

Respiratory Distress as an Isolated Manifestation of Status Epilepticus

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Abstract

Respiratory distress by laryngospasm as an isolated manifestation of epileptic seizure is rare phenomenon. We report on a 1-month-old infant with recurrent respiratory distresses by laryngospasm as an isolated manifestation of status epilepticus and resultant periarrests, while being monitored in our intensive care unit. This case of an atypical manifestation of status epilepticus highlights the need for increased index of suspicion for epileptic seizure in patients with recurrent respiratory distresses and demonstrates that respiratory distress such as laryngospasm may represent another potential cause of sudden unexpected death in epilepsy.

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Keywords

- Seizures
- Status epilepticus
- Respiratory distress
- Laryngospasm
- Sudden unexpected death in epilepsy (SUDEP)

ABBREVIATIONS

EEG: Electroencephalogram; SUDEP: Sudden Unexpected Death in Epilepsy

CASE PRESENTATION

A 1-month-old boy with no perinatal problems presented with severe cough and sleep disturbance for 3 days. Chest auscultation revealed fine crackles bilaterally and a chest X-ray showed coarsening of interstitial marking in both mid lung fields (Figure 1). He was diagnosed with bronchiolitis and admitted. The next day, he had a seizure. Seizure type was generalized tonic-clonic. When the ward staff arrived, he presented with repetitive jerks of both arms and legs. Furthermore, leftward conjugate eye deviation, drooling, and cyanosis were observed. The attack discontinued after administration of intravenous lorazepam. Seizure duration was 20 minutes. Chest auscultation revealed decreased breath sound and extreme wheezing aggravated relative to the previous day. He was transferred to the intensive care unit where he was intubated and placed on mechanical ventilation. About 2 hours later, he developed bradycardia with cyanosis. Chest auscultation revealed extremely decreased breath sound, and paradoxical chest and abdominal movements were observed. We did not observe any motor symptoms of seizure. A presumptive diagnosis of bronchiolitis obliterans was made and he was resuscitated by administration of intravenous epinephrine, intravenous hydrocortisone, and continuous nebulized salbutamol. After restoration, several periarrests preceded by cyanosis occurred as described above. We performed bedside Electroencephalogram (EEG) monitoring. EEG showed periodic polyspikes on the left fronto-centro-parietal region that occurred simultaneously to the cyanotic attacks (Figure 2). Levels of serum electrolytes, including calcium and magnesium, were normal and repeated chest X-ray showed little change. Based on the diagnosis of status epilepticus, intravenous phenobarbital was administrated at loading dose over 10 minutes; this resulted in cessation of the patient's respiratory distress and periarrests. Brain magnetic resonance imaging was also performed; no abnormalities were detected. There were no additional seizure attacks and he was transferred to the general ward and observed for a next few days. Oral phenobarbital was prescribed and treatment for bronchiolitis continued. The patient had recovered to baseline status at the 1-monthfollow-up and his EEG findings had normalized.

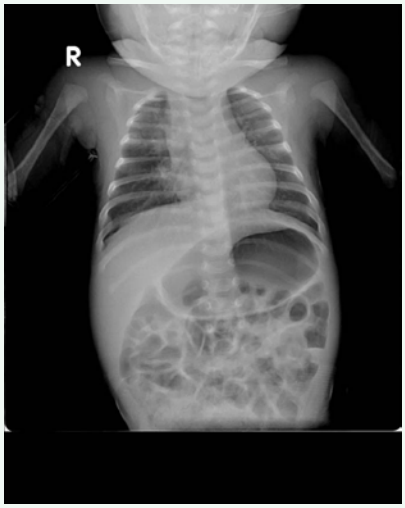

Figure 1 Chest radiograph showed coarsening of interstitial markings on the bilateral medial lung fields and the ileus.

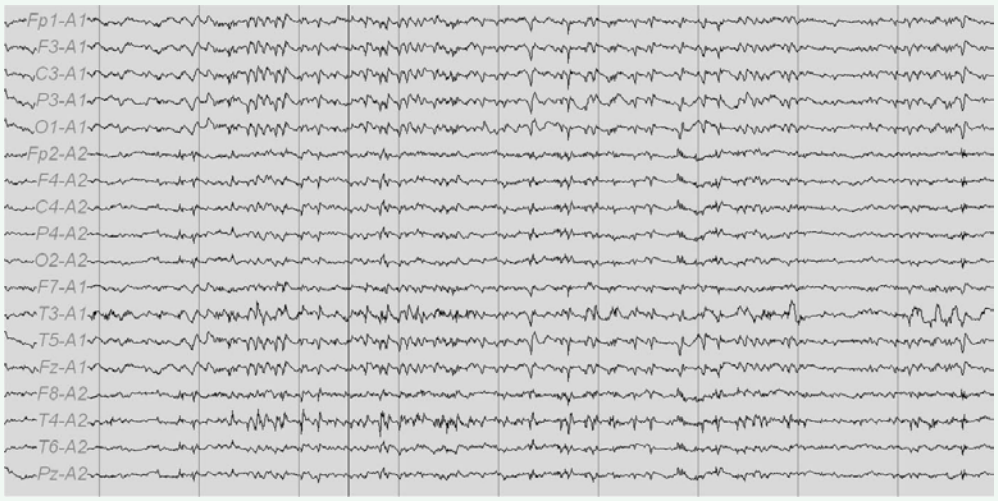

Figure 2 Electroencephalogram recorded during respiratory distress showed periodic polyspikes on the left fronto-centro-parietal region.

DISCUSSION

To our knowledge, only four reports have described laryngospasm as an isolated manifestation of epileptic seizure [1-4]. While our patient developed recurrent cyanosis with periarrests, chest auscultation revealed total airway obstruction, and paradoxical chest and abdominal movements were observed. These findings implied upper airway obstruction such as laryngospasm, but the patient was misdiagnosed with bronchiolitis obliterans. However, the laryngospasm was due to epileptic seizure, because EEG monitoring revealed ictal patterns and recurrent laryngospasm discontinued after administration of phenobarbital. Nevertheless, the initial symptoms and signs of our patient also implied co-morbidity of brochiolitis. The larynx is served by the 10th cranial nerve via the superior and interior laryngeal nerve branches. A number of vital laryngeal reflexes exist. Laryngeal spasm, bronchial constriction, slowing of heart rate, and apnea can have life-threatening effects on the cardiovascular and respiratory systems, and are known to be mediated by laryngeal reflexes. However, the pathophysiology of epileptic stimulation is not known [1,2]. Meanwhile, the pathophysiology of sudden unexpected death in epilepsy (SUDEP) is unclear. Certain risk factors for SUDEP have been identified, and a small number of witnessed cases of SUDEP have been reported, but only a handful has occurred in the setting of continuous EEG monitoring. Suspected mechanisms include cardiac arrhythmia, respiratory arrest, and primary cessation of brain activity. Some authors reported that seizures resulted in laryngospasm, which is one of multiple probable mechanisms of SUDEP [5,6]. This is the first report of a case of status epilepticus presenting as isolated laryngospasm. This case highlights the need for increased suspicion of epileptic seizure in patients who present with recurrent respiratory arrest, and suggests that respiratory arrest symptoms, such as laryngospasm, may be a potential cause of SUDEP.

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