Thoracocervicofacial purpura and hypoxemia after generalized tonic-clonic seizure

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Summary

We report a case of a 41-year-old man who showed thoracocervicofacial purpura after generalized tonic-clonic convulsion. The patient also had hypoxemia with chest CT findings of non-homogeneous high density in the posterior areas in both lungs, without signs of infection or heart failure. The purpura suggested that strong valsalva maneuver during airway closure and the resultant increase in systemic arterial and pulmonary vascular pressure may have led to pulmonary congestion.
**Introduction**

Thoracocervicofacial purpura is a rare consequence after epilepsy, and has been speculated to be caused by strong valsalva maneuver during tonic-clonic seizure [1, 2]. We describe a patient who showed thoracocervicofacial purpura and hypoxemia after tonic-clonic seizure. Hypoxemia without infection is a potential and important symptom following this type of epilepsy.

**Case Report**

A 41-year-old man with a past history of epilepsy until the age of 3 fell on the floor in his office with generalized tonic-clonic seizure. He had transient narrowing of the visual field 2 hours before the seizure, and was transferred to our hospital. On admission, a physical examination showed blood pressure of 127/49 mmHg, heart rate 120 beats per minute, temperature 36.3 °C, and respiratory rate 18 per minute. The oxygen saturation was 92% in room air. Purpura was observed in the thoracocervicofacial area, especially marked around the palpebrae (Fig. 1). His neurological examination was unremarkable. Routine laboratory data demonstrated white blood cell

![Figure 1. On admission, the patient presented with purpura in the thoracocervicofacial area, especially marked around the palpebrae. The patient was also hypoxemic requiring oxygen inhalation.](image)
count 13,070/mm$^3$ (neutrophil 38.8%, lymph 52.7%), platelet count 283,000/mm$^3$, creatine kinase 261 IU/L (normal range: 12-142 IU/L), and C-reactive protein 0.1 mg/dL (normal range, <0.3 mg/dL). Coagulation test results including prothrombin time, activated partial prothrombin time and fibrinogen level were normal. An electrocardiogram (EKG) revealed sinus tachycardia. MRI of the head showed no abnormality. Chest X ray showed no abnormality in pulmonary fields and no enlarged heart. CT of the chest demonstrated non-homogenous density increase in the posterior areas adjacent to the pleura in both lungs (Fig. 2). An electroencephalogram showed high-amplitude delta bursts at 2.5-3 Hz in the frontal area during hyperventilation load. The patient was observed in the hospital without antibiotic treatment. Examinations 24 hours later showed oxygen saturation of 97% when breathing room air, white blood cell count 9080 /mm$^3$, and creatine kinase 1191 IU/L. The purpura did not worsen. The patient was started on antiepileptic treatment and was discharged. At the follow-up 15 days later, the purpura had disappeared, white blood cell count was normalized, and the abnormal appearance of the lungs on CT was resolved.

**Figure 2.** A CT image obtained on admission reveals high signal intensity in the posterior areas adjacent to the pleura in both lungs.
Discussion

It is known that the valsalva maneuver may result in a pronounced elevation of mean arterial pressure leading to complications including intracranial bleeding [3, 4], acute aortic dissection [5], and thoracocervicofacial purpura as was observed in our patient [1, 2]. This phenomenon has been reported to occur under heavy resistance exercises in which the breath is held, e.g., weight lifting, swimming [3-5]. Thoracocervicofacial purpura rarely occurs after epileptic seizure and has been speculated to be caused by an acute increase in intrathoracic pressure with the glottis closed, as in the valsalva maneuver [1, 2]. This sign is important because it is suggestive of general tonic-clonic seizure even though the seizure is not witnessed.

Our patient had another important complication, afebrile hypoxemia. Decreased oxyhemoglobin saturation was sustained several hours after seizure, and it resolved within 24 hours without the administration of antibiotics. There was no enlarged heart, and CT showed increased density in the posterior areas in both lungs without findings of pneumonia, suggesting mild acute respiratory distress syndrome [6]. C-reactive protein was within the normal range. There was peripheral blood leukocytosis, but it has been reported that nearly 50% of children with afebrile seizure show peripheral blood leukocytosis without bacterial infection [7]. Based on these findings, it is probable that the hypoxemia of our patient was not due to respiratory infection.

Ictal hypoxemia has been studied in the context of sudden unexpected death in epilepsy (SUDEP), and a recent study has reported that central apnea or hypopnea occur in 50 of 100 seizures [8]. However, ictal hypopnea was not a cause of the sustained hypoxemia. Seizure-related heart failure is another pathomechanism leading to hypoxemia [9], but our patient did not show evidence of heart failure. Pulmonary congestion or edema has been reported to be a possible cause of SUDEP [10, 11] and estimated to result from an increase in sympathetic drive with intense vasoconstriction and increased systemic arterial and pulmonary vascular pressure [10, 12]. As was suggested by the purpura, our patient had a strong valsalva maneuver during seizure, and the consequent marked increase in systemic arterial and pulmonary vascular pressure may have induced pulmonary congestion or edema. Thus strong Valsalva maneuver may cause postictal hypoxemia, although it is uncertain whether it could lead to SUDEP.

This case highlighted that sustained hypoxemia and thoracocervicofacial purpura may follow generalized tonic-clonic seizure because of the accompanying strong Valsalva maneuver.

References


