Milan Sova[1], David Franc[2], Filip Ctvrtlik[3], Petr Jakubec[1], Amjad Ghazal Asswad[4], Vitezslav Kolek[1]

[1]Department of Respiratory Medicine, Faculty of Medicine and Dentistry, Palacky University and University Hospital Olomouc, Czech Republic
[2]Department of Neurology, Faculty of Medicine and Dentistry, Palacky University and University Hospital Olomouc, Czech Republic
[3]Department of Radiology, Faculty of Medicine and Dentistry, Palacky University and University Hospital Olomouc, Czech Republic
[4]Palacky University Olomouc, Czech Republic

Neurogenic pulmonary oedema as a rare complication of epileptic seizures

Abstract

Introduction: Neurogenic pulmonary oedema (NPE) is a very rare complication of epileptic seizures, which could potentially increase mortality.

Material and methods: The case of a 66-year-old male patient with NPE caused by repeated epileptic seizures is reported. Rapid resolution of pulmonary oedema is well documented by X-ray and computed tomography images.

Conclusions: Neurogenic pulmonary oedema could potentially increase mortality, and thus, it is important to perform a chest X-ray in all patients presenting with seizures and dyspnoea.

Key words: neurogenic pulmonary oedema, epilepsy, seizure

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Introduction

Neurogenic pulmonary oedema (NPE) is a clinical syndrome characterised by the acute onset of pulmonary oedema following a central nervous system (CNS) insult [1]. Although its aetiology is not completely understood, it probably results from a catecholamine surge which can be caused by many CNS events like spinal cord injury, subarachnoid haemorrhage, meningitis, subdural haemorrhage and status epilepticus [2]. It was first described in 1908 by Shanahan in 7 patients with status epilepticus [3]. Two series of patients with NPE were also published based on head wounds sustained by soldiers during World War I [4] and the Vietnam War [5]. The occurrence of NPE in patients with epilepsy without status epilepticus is rare, with only a few such cases having been published. In this case report, we present such a case of a patient with fully developed NPE following epileptic seizures, but without the development of status epilepticus.

Material and methods

The patient was a 66-year-old male with a known history of temporal lobe epilepsy and two prior epileptic seizures in July and September 2013. He was taking levetiracetam 2000 mg/day. On the 2nd of September 2014, his spouse noticed three epileptic seizures (3–5 minutes long) with generalised tonic-clonic convulsions, bit tongue and post-paroxysmal fuzziness. The patient was transported to the emergency department. The neurological examination did not find any neurological pathology besides mild fuzziness. There was no aphasia, paresis, restlessness, aggressiveness or other behavioural or thinking disorders. He suffered a fourth seizure shortly after admission, which was again with tonic-clonic convulsions, though the beginning of the seizure was not seen.

After admission, a CT scan of the brain was performed which was negative. Hypoxaemia during transport (90% SpO_2 on air) meant that further diagnostic procedures were required.

Address for correspondence: Milan Sova, Department of Respiratory Medicine Faculty of Medicine and Dentistry, Palacky University and University Hospital Olomouc, Czech Republic; e-mail: milan.sova@email.cz
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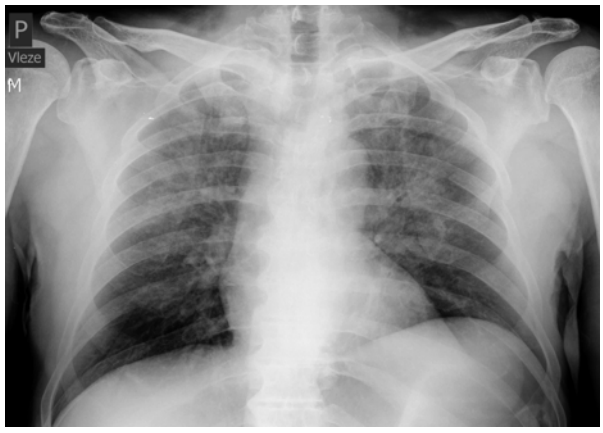

Figure 1. Nonhomogeneous opacities including upper parts of the lung bilaterally

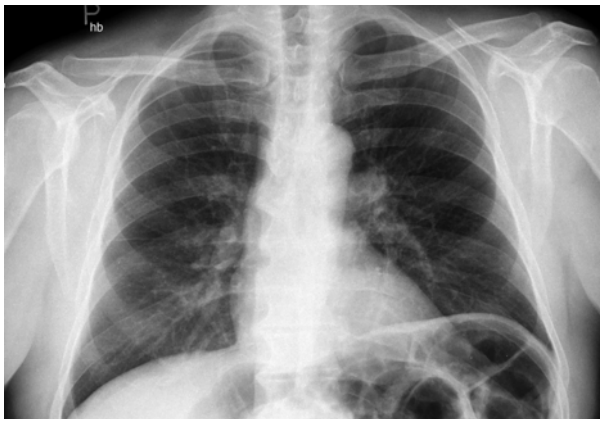

Figure 3. Chest X-ray 35 hours after initial examination — complete regression of opacities

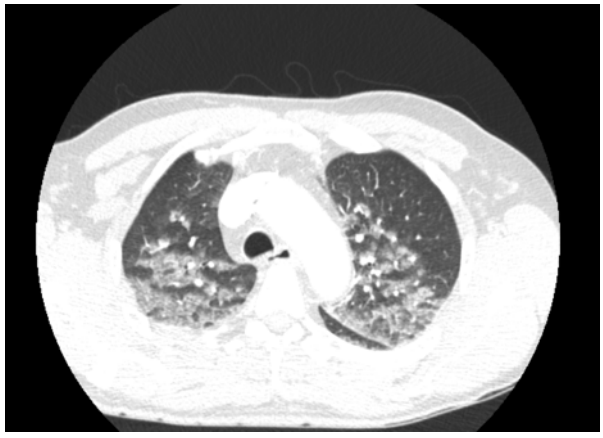

Figure 2. Thorax CT scan — ground glass opacities, bilaterally, predominantly dorsally

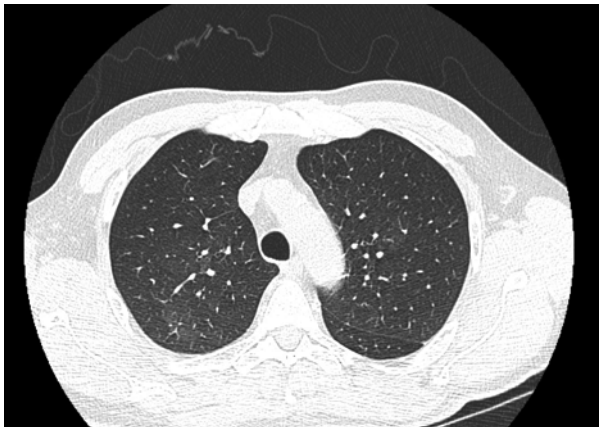

Figure 4. Thorax high-resolution CT scan 55 hours after the initial examination

All biochemical results were normal, including C-reactive protein (CRP) level. Complete blood count results were also in the physiological range. A baseline chest X-ray is shown in Figure 1. Nonhomogenous opacities were found bilaterally.

Negative CRP together with a normal leukocyte count ruled out the suspicion of pneumonia. Hypoxaemia was present (PaO_2 = 7.8 kPa) with normocapnia ($PaCO_2$ = 6.0 kPa). For further evaluation, a chest CT was indicated. This CT scan is shown in Figure 2.

The patient was indicated hospitalisation at the Department of Neurology; he remained seizure- free throughout its duration. However, due to unclear pulmonary findings, the man was transferred to the Department of Respiratory Medicine the next day. An initial chest X-ray following this transfer is shown in Figure 3.

For further clarification of the X-ray findings, a high-resolution chest CT scan was performed. The result is shown in Figure 4.

For more precise diagnosis, bronchoscopy with bronchoalveolar lavage (BAL) was performed. Small amounts of blood were found in the airways, with the BAL fluid being haemorrhagic. An elevation in D-dimers prompted a ventilation/ /perfusion lung scan to be performed which was negative, alleviating the suspicions of a pulmonary artery embolism. During his hospitalisation at the Department of Respiratory Medicine, the patient was free of seizures or other clinically relevant events. During follow-up (until 2/2019), similar epileptic seizures have not reoccurred.

Discussion

Neurogenic pulmonary oedema is rare in patients without known status epilepticus seizures. Pathogenesis is not completely understood. Currently, several clinicopathologic paradigms have been proposed to explain the clinical syndrome of NPE. These are the following:

— Neurocardiac NPE (neurologic insult leads to direct myocardial injury and subsequently causes Takotsubo cardiomyopathy with pulmonary oedema).
— Neuro-haemodynamic NPE (ventricular compliance is indirectly altered by an abrupt increase in systemic and pulmonary pressures following CNS injury).
— Blast theory (severe abrupt increases in systemic and pulmonary pressures following the catecholamine surge result in a net shift of blood volume from the systemic circulation to the low resistance pulmonary circulation).
— Pulmonary venule adrenergic hypersensitivity (massive sympathetic discharge following CNS injury directly affects the pulmonary vascular bed, and oedema develops regardless of any systemic changes).
— The recommended therapeutic options are also not very clear. It is important to treat the underlying neurological condition which caused the NPE. In animal models, there were successful findings using α-adrenergic blocking agents [6–8], β-adrenergic blocking agents [6, 7] as well as some other therapeutic approaches. In this case, only antiepileptic medication together with oxygen therapy was used.

Conclusions

Neurogenic pulmonary oedema is a rare complication of epileptic seizures which could worsen prognosis and increase mortality and morbidity [9]. For its diagnosis, it is necessary to perform a chest X-ray or chest CT scan. The recommended treatment is not well established but it seems that the use of α or β adrenergic blocking agents may be useful.

Author contributions

All authors contributed equally to the preparation of this manuscript.

Conflict of interest

None of the authors have any conflicts of interest to declare regarding this manuscript.

References:

1. Davison DL, Terek M, Chawla LS. Neurogenic pulmonary edema. Crit Care. 2012; 16(2): 212, doi: 10.1186/cc11226, indexed in Pubmed: 22429697.
2. Fontes RBV, Aguiar PH, Zanetti MV, et al. Acute neurogenic pulmonary edema: case reports and literature review. J Neurosurg Anesthesiol. 2003; 15(2): 144–150, indexed in Pubmed: 12658001.
3. Shanahan W. Acute pulmonary edema as a complication of epileptic seizures. NY Med J. 1908; 37: 54–56.
4. Moutier F. Hypertension et mort par oedeme pulmo aigu chez les blesses cranio-encephaliques. Presse Med. 1918; 26: 108–109.
5. Simmons RL, Heisterkamp CA, Collins JA, et al. Respiratory insufficiency in combat casualties. 3. Arterial hypoxemia after wounding. Ann Surg. 1969; 170(1): 45–52, doi: 10.1097/00000658-196907000-00006, indexed in Pubmed: 5789529.
6. Brashear RE, Ross JC. Hemodynamic effects of elevated cerebrospinal fluid pressure: alterations with adrenergic blockade. J Clin Invest. 1970; 49(7): 1324–1333, doi: 10.1172/JCI106348, indexed in Pubmed: 4393489.
7. Malik AB, Minnear FL, Malik AB. Mechanisms of neurogenic pulmonary edema. Ann N Y Acad Sci. 1982; 384(1): 169–190, doi: 10.1111/j.1749-6632.1982.tb21371.x, indexed in Pubmed: 6953818.
8. Nathan MA, Reis DJ. Fulminating arterial hypertension with pulmonary edema from release of adrenomedullary catecholamines after lesions of the anterior hypothalamus in the rat. Circ Res. 1975; 37(2): 226–235, doi: 10.1161/01.res.37.2.226, indexed in Pubmed: 1149197.
9. Nascimento FA, Tseng ZH, Palmiere C, et al. Pulmonary and cardiac pathology in sudden unexpected death in epilepsy (SUDEP). Epilepsy Behav. 2017; 73: 119–125, doi: 10.1016/j.yebeh.2017.05.013, indexed in Pubmed: 28633090.