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INTRODUCTION

We present the case of a 38-year-old lady with no prior history of cardiac disease who experienced sudden onset chest pain and acute left ventricular failure 4 hours following explantation of stereoelectroencephalogram electrodes. Echocardiography demonstrated hypokinesis of the cardiac apex and septum consistent with Takotsubo stress cardiomyopathy which has not been previously reported in the context of stereoelectroencephalography. The aim of this case report was to raise awareness of the potential complication of Takotsubo stress cardiomyopathy in an epilepsy surgery program.
CASE REPORT

A 38-year-old, right-handed lady developed chest pain and acute pulmonary edema 4 hours following explantation of stereoencephalogram (sEEG) electrodes. She had poorly controlled, post-traumatic right temporal lobe epilepsy but no additional past medical history or risk factors for cardiovascular disease. MRI brain demonstrated multiple areas of gliosis, particularly laterally and anteromedially in the right temporal lobe, the right, and to a lesser degree, left orbitofrontal cortex, with focal white matter loss in the corpus callosum. Video-telemetry was consistent with focal-onset, nondominant mesial temporal or orbitofrontal lobe seizures; semiology consisted of right upper limb and oral motor automatisms preceded by prolonged aura, including olfactory and autonomic features. Generalized convulsive seizures were not observed during this period of recording. Robotic sEEG (Renishaw Neuramate) was undertaken to further localize seizure onset; 13 electrodes were implanted in the right hemisphere in a frontotemporal distribution and 1 in the left orbitofrontal region (Figure 1A,B). Routine postinsertion CT demonstrated a small parenchymal hematoma related to the right posterior temporal electrode ([electrode F]; entry via middle temporal gyrus, deep contacts in the fusiform gyrus) which was asymptomatic. Over 9 days, a total of 14 stereotyped seizures were recorded (Figure 1C); all initiated from the right mesial temporal structures starting in the amygdala with rapid spread to the hippocampus and right temporal pole. Apparently uneventful explantation occurred at day 10 on the ward. This was several hours after the last recorded seizure, and prophylactic enoxaparin had been withheld for 48 hours.

Four hours postexplantation during which time seizures did not occur, the patient experienced severe central chest pain at rest with radiation to the neck and associated dyspnea, diaphoresis, and nausea. Pain responded to glyceryl trinitrate and morphine. She denied headache. Oxygen (O2) saturations were 73% on air, improving to 98% with 3 L O2. There was hemodynamic instability, sinus tachycardia (rate 125/min) and episodic hypotension (minimum systolic 70 mmHg). Cardiorespiratory examination was consistent with pulmonary edema secondary to acute left ventricular (LV) failure, and there were no focal neurological findings or seizures. Electrocardiogram demonstrated new-onset anterolateral T-wave inversion with reciprocal changes in leads II, III, and aVF (Figure 2A,B). Chest X-ray findings confirmed pulmonary edema (Figure 2C). Intravenous frusemide was administered, and, given concern regarding the possibility of non-ST elevation myocardial infarction (NSTEMI), dual antiplatelet therapy was commenced.

Clinical improvement with resolution of chest pain and dyspnea occurred over 24 hours. Troponin level rose from 13 to 33 ng/L. ECG changes resolved, and the coronary angiogram was normal. Antiplatelet therapy was discontinued at 48 hours. Transthoracic echocardiogram revealed evidence of moderate LV systolic dysfunction (estimated ejection fraction 40%–45%) and hypokinesis of the cardiac apex as well as the anterior and anteroseptum. The right temporal lobe hematoma was again noted on MRI brain at 2 days postexplantation along with right frontal pole edema (Figure 3A,B) but there was no extension of the hematoma when compared with the initial postexplantation CT scan. TTE demonstrated improvement in LV systolic function at 4 weeks and near normal function at 6 months.

Following initial concern regarding the possibility of NSTEMI, the diagnosis was revised to takotsubo stress cardiomyopathy (TTS). TTS, also known as “stress-induced cardiomyopathy” and “broken heart syndrome,” is characterized by LV dysfunction with regional wall abnormalities including apical ballooning and focal wall-motion abnormalities; the end-systole radiological appearance of the left ventricle is said to resemble a Japanese octopus trap (“takotsubo”). TTS occurs more often in postmenopausal females and at times of emotional and physical stress. The pathogenesis remains uncertain although sympathetic stimulation is key and a link between the insular cortex and central autonomic network recognized. Acute neurological diseases including subarachnoid hemorrhage, stroke, and seizures are well-recognized precipitants for TTS, and an association between TTS and sudden unexplained death in epilepsy (SUDEP) has been proposed. TTS has not previously been recorded in the context of sEEG.

Level C evidence for treatment of TTS is available, and management is generally based on guidelines for treatment of acute coronary artery syndrome with particular consideration given to beta-blockers in view of association with elevated catecholamine levels, diuretics for pulmonary edema, and nitroglycerin in LV failure.

Intracranial hemorrhage (ICH) is well recognized to be a precipitant for TTS, and ICH has been reported to occur in approximately 1% patients undergoing sEEG. The presence of temporal lobe hemorrhage could therefore have been a
trigger for TTS. Takosubo syndrome has also been reported several days after convulsive and nonconvulsive seizures, so seizure activity may also have been a contributory factor. While we cannot categorically state that explantation induced TTS in our patient, we consider the timing suggestive although the association has not, to the best of our knowledge, been reported previously. Furthermore, the multiple potential triggers may have contributed to the presentation.

Given the potential for overlapping risk factors of seizures and ICH, this case highlights the importance of awareness of TTS in the context of an epilepsy surgery program. We advise early involvement of cardiologists in the care of patients suspected of having TTS and, if clinically safe to do so, prompt neuroimaging to exclude ICH.

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FIGURE 2  ECG changes and chest X-ray during presentation with acute chest pain. ECGs on admission (A) and during the acute episode (B) demonstrate new anterolateral T wave inversion with reciprocal changes in leads II, III and aVF. The chest X-ray (C) appearances are consistent with pulmonary edema.
CONFLICTS OF INTEREST
None of the authors has any conflict of interest to declare. We confirm that we have read the Journal’s position of issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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