Case report

A case report of cerebral arterial gas embolism (CAGE) associated with Takotsubo cardiomyopathy

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Abstract

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A 43-year-old female scuba diver was retrieved and treated following a rapid ascent and presumed cerebral arterial gas embolism (CAGE). She subsequently developed respiratory distress and was found to have Takotsubo cardiomyopathy, with transient left ventricular dysfunction, elevated cardiac enzymes, and normal CT coronary angiography. We believe this to be the first report of CAGE associated with Takotsubo cardiomyopathy.

Introduction

Pulmonary barotrauma (PBT) with cerebral arterial gas embolism (CAGE) is a known risk of scuba diving. Respiratory compromise following CAGE is common, and is often a result of non-fatal drowning in the disabled diver.[1] Gas embolisation of the coronary circulation is also described, and can cause cardiac dysfunction.[2]

Takotsubo cardiomyopathy (TCM) is a pattern of cardiac dysfunction seen in situations following high sympathetic stimulation such as subarachnoid haemorrhage and ischaemic stroke.[3,4] It may also occur in otherwise uneventful diving, presenting as immersion pulmonary oedema.[5] TCM is described in a case of iatrogenic gas embolization,[6] but has not previously been described in divers suffering from CAGE.

We present a case of a diver suffering from CAGE who developed severe pulmonary oedema requiring intubation and mechanical ventilation, secondary to TCM.

Case report

A previously well 43-year-old female diver on the first dive of the day developed mask problems at the beginning of the dive, and ascended rapidly from a depth of 17 metres' sea water (msw). She was symptom free at depth but lost consciousness on surfacing. Her dive instructor observed her to be blue and limp, inflated her buoyancy compensator and retrieved her to the boat. She remained unresponsive for up to 10 minutes.

After regaining consciousness she vomited, but displayed no gross neurological abnormalities. She was coughing with mild shortness of breath, but no chest pain. Helicopter retrieval to the nearest hospital was arranged, with her initial management consisting of high flow oxygen. In view of the likely diagnosis of CAGE she was kept supine. She was noted at the time of retrieval to have bilateral respiratory crepitations, peripheral oxygen saturation (SpO_2) of 95% on 8 L·min^{-1} of oxygen via a non-rebreather face mask, and an elevated respiratory rate (number of breaths per minute not recorded).

On arrival at hospital, approximately two hours after surfacing from the dive, she had a Glasgow Coma Score (GCS) of 15, and no focal neurology or evidence of pulmonary barotrauma. She had a tachycardia of 120 beats per minute, a respiratory rate of 35–40 breaths per minute, and required supplemental oxygen to maintain the SpO_2 above 95%. Blood tests demonstrated haemoconcentration, a metabolic acidosis, and a PaO_2:FiO_2 (P:F) ratio of 155 (normal around 500). A chest X-ray showed bilateral alveolar infiltrates that were considered to be in keeping with salt water aspiration, but no radiological evidence of pulmonary

barotrauma (e.g., pneumothorax). An ECG demonstrated a sinus tachycardia with no acute ST segment elevation or Q waves to suggest a myocardial infarction. A provisional diagnosis of resolving CAGE with salt water aspiration was made, and transfer to our centre for hyperbaric oxygen therapy was arranged. One litre of intravenous crystalloid was administered, and high flow oxygen continued.

On arrival for recompression she remained neurologically intact with a sinus tachycardia of 128 bpm, blood pressure 127/87 mmHg, an SpO_2 of 98% on 15 $L \cdot min^{-1}$ of oxygen. Her ECG was unchanged. Widespread crepitations were noted on chest auscultation. In view of the risk of further embolization by residual gas trapped in the pulmonary veins or heart chambers, or neurological deterioration, she was recompressed with an RN62 recompression table (US Navy Treatment Table 6). She received oxygen via a hood system.

During the hyperbaric treatment she developed increasing respiratory distress and diaphoresis, with desaturation during the air breaks. There was no chest pain. Towards the end of the treatment, the patient was expectorating pink frothy sputum. On completion of recompression she required intubation and ventilation for progressive hypoxaemia. Pulmonary oedema fluid was noted in the endotracheal tube. Repeat chest X-ray again demonstrated bilateral fluffy opacification, and there continued to be no radiological evidence of pneumothorax. Flexible bronchoscopy showed the endotracheal tube to be in good position, a small amount of clear watery secretions in the trachea, but no evidence of aspiration or segmental collapse.

After arrival in the intensive care unit (ICU), transthoracic echocardiography (TTE) showed global left ventricular dysfunction with an ejection fraction (EF) of 20–25% with mid-ventricular akinesia. The heart was otherwise structurally normal, with no evidence of outflow obstruction. The appearance was consistent with a mid-ventricular stress cardiomyopathy. The ECG now showed an inverted T wave in lead I, and a flattened T wave in V6. The T waves were biphasic in leads II, III, and aVF. There was no ST segment elevation. Sinus tachycardia and hypotension were treated with a small fluid bolus and vasopressors. Low dose milrinone was added empirically to improve ventricular function. The patient remained sedated, intubated and ventilated for 36 hours. During this time she remained neurologically stable. Her sedation was ceased and she responded to verbal commands with no evidence of focal neurological deficit and a P:F ratio over 300. She was extubated with an SpO_2 of 98% (inspired oxygen fraction 30%). Bedside TTE prior to extubation revealed a substantial improvement of the LV dysfunction with visual estimate of EF of 45–50% (at around 34 hrs after ICU admission, and 48 hrs after the dive).

The patient's Troponin I peaked at 2956 $ng \cdot L^{-1}$ (normal < 10). Computed tomography coronary angiography demonstrated a coronary calcium score of 0. The coronary circulation was normal other than an aberrant origin of the right coronary artery (RCA); a slit-like origin from the ascending aorta above the sino-tubular junction on the left side, superior to the origin of the left coronary artery. The initial course of the RCA ran between the aorta and the main pulmonary artery. The left anterior descending (LAD) artery was described as a type III vessel with no disease and no myocardial bridging. Exercise stress testing was performed 10 days after the dive, with the patient exercising to 9.7 metabolic equivalents (MET) with no ECG changes or chest pain. The test was stopped due to dyspnoea and dizziness. The right ventricle was normal in size and function on multiple TTEs over the course of the patient's hospital admission. Magnetic resonance imaging (MRI) of the brain showed evidence of a small infarction in the right post-central sulcus consistent with the diagnosis of CAGE, but no other abnormalities. The patient denied experiencing chest pain at any time before or during her admission.

At follow-up three weeks following the dive she was completely well with resolution of all symptoms. Repeat TTE at this time showed normal left ventricular systolic function with an EF of 67%.

Discussion

Given this diver's history of a rapid ascent, loss of consciousness on surfacing and brain MRI evidence of infarction, it is likely that the primary pathology in this case was pulmonary barotrauma leading to CAGE.

The typical natural history of CAGE is for a sudden onset of neurological abnormality followed by a partial or complete recovery. Relapse can follow due to re-embolisation by gas still in the pulmonary veins or cardiac chambers, or due to inflammatory changes in the injured brain and cerebral vessels. Initial recovery is primarily due to the redistribution of bubbles through the cerebral circulation, in part due to a hypertensive response to the brain injury.[7–9]

Takotsubo cardiomyopathy (TCM) was first described in 1991, and is often characterized as 'stress cardiomyopathy'.[10] The commonest (but not only) form is apical ballooning of the left ventricle from which the term 'takotsubo' takes its name. The Mayo Clinic criteria specify, as one of the criteria, "*transient hypokinesis, akinesis, or dyskinesis in the left ventricular mid segments with or without apical involvement; regional wall motion abnormalities that extend beyond that expected from a single epicardial vascular distribution*".[10] Isolated mid-ventricular and basal LV dysfunction / isolated mid-ventricular LV dysfunction (apical-sparing TC) is a described variant.[11] Stress cardiomyopathy may be found in approximately 2% of patients presenting with probable myocardial infarction, however it has a relatively low mortality of 1–2%, and recurrence is less than 1%. The average clinical recovery time is approximately 18 days. The condition occurs most commonly in older individuals with a high female

predominance. The aetiology is unknown. It often follows a severe emotional shock although many triggering events have been reported. Stress cardiomyopathy is diagnosed first by suspicion, often in the context of apparent coronary artery disease. Echocardiographic findings are regional hypokinesia or akinesia, predominantly in the left ventricle (although the right ventricle can be affected). The extent of the regional ventricular muscle dysfunction is usually outside expected coronary artery territory, and the diagnosis should be confirmed by angiography showing no significant coronary disease. It may be accompanied by ECG changes of ST elevation and/or a significant troponin rise. During the recovery phase, the use of beta blockers and afterload reduction is considered beneficial.

The precipitation of TCM following non-diving intracerebral events such as subarachnoid haemorrhage and ischaemic stroke is described.[3,4] Subarachnoid haemorrhage in particular is associated with an extreme sympathetic response with high levels of circulating catecholamines, which is one of the proposed mechanisms of the myocardial injury in TCM.[10,11]

We were able to find a single case in the literature of a case of TCM which was precipitated by iatrogenic CAGE whilst undergoing a computed tomography-guided transthoracic needle biopsy.[6] This case demonstrated a typical pattern of TCM with apical ballooning, raised cardiac enzymes and normal coronary angiography. The ventricular dysfunction resolved five days following the event.

Villela et al. reported a case of a previously well 34-year-old diver who had a likely CAGE, with troponin rise, akinesis of the apex of the left ventricle and hypokinesis of the mid infero-septal segment, an EF of 50% and normal coronary angiography.[12] Two weeks later, the regional wall motion abnormalities and ventricular function had normalised. Their case differs from ours in that their patient reported significant chest pain responding to vasodilator therapy, and the development of pericarditis. The authors suggested the pathology was likely to be coronary artery gas embolism, with TCM a possible alternative.

The differential diagnosis of respiratory failure in our case includes immersion pulmonary oedema, coronary gas embolism, or aspiration with non-fatal drowning. Non-fatal drowning does not explain the severe cardiac dysfunction, unless as a separate cause of TCM.[13] Coronary gas embolism does not explain the pattern of myocardial dysfunction seen on echocardiography. Nor is it explained by occlusion of the RCA at or near its origin. Immersion pulmonary oedema is also unlikely as the respiratory symptoms were not present at depth, the water was warm (27–29 degrees Celsius), and the dive did not involve severe exertion.[14] The respiratory symptoms were absent at depth, worsened following exiting the water and further during recompression, which is more in keeping with pulmonary oedema due to left ventricular failure. The nature of the deterioration during the course of the recompression may relate to the increased afterload for the left heart due to systemic vasoconstriction. Alternatively, it may have represented pulmonary oxygen toxicity superimposed on already impaired respiratory function.

There are a number of reports and case series of cardiomyopathy developing in seemingly uneventful dives, and this may represent a subset of divers who develop immersion pulmonary oedema, although the exact mechanism is unclear.[14–17]

Conclusion

We believe this case is the first to describe Takotsubo cardiomyopathy in a diver suffering from CAGE. We think the diagnosis should be considered in divers suffering from CAGE who develop respiratory distress or cardiovascular instability.

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Errata

In a review article (Hoencamp E, van Dongen TTCF, van Ooij PJAM, Wingelaar TT, Vervelde ML, Koch DAA, van Hulst RA, Hoencamp R. Systematic review on the effects of medication under hyperbaric conditions: consequences for the diver. Diving and Hyperbaric Medicine. 2019 June 30;49(2):127–136. doi: 10.28920/dhm49.2.127-136. PMID: 31177519.) Table 1 was labelled twice, meaning that the subsequent tables were labelled incorrectly. The correct table labelling now appears in our electronic version online for download by Society members, and will be correct in PubMed Central®'s full text version. The error will persist if accessed through Elsevier, Clarivate, the National Library of Australia and in the small number of print copies distributed.