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SEVERE CAPILLARY LEAK SYNDROME AFTER INNER EAR DECOMPRESSION SICKNESS IN A RECREATIONAL SCUBA DIVER

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☐ **Abstract—Background: Post-decompression shock with plasma volume deficit is a very rare event that has been observed under extreme conditions of hypobaric and hyperbaric exposure in aviators and professional divers. Case Report: We report a case of severe hypovolemic shock due to extravasation of plasma in a recreational scuba diver presenting with inner ear decompression sickness. Impaired endothelial function can lead to capillary leak with hemoconcentration and hypotension in severe cases. This report suggests that decompression-induced circulating bubbles may have triggered the endothelial damage, activating the classic inflammatory pathway of increased vascular permeability. Conclusion: This observation highlights the need for an accurate diagnosis of this potentially life-threatening condition at the initial presentation in the Emergency Department after a diving-related injury. An elevated hematocrit in a diver should raise the suspicion for the potential development of capillary leak syndrome requiring specific treatment using albumin infusion as primary fluid replacement.** © 2013 Elsevier Inc.

☐ **Keywords—diving; decompression sickness; capillary leak; hypovolemic shock**

INTRODUCTION

Decompression sickness (DCS) is an acute clinical disorder affecting scuba divers that results from the excessive formation of circulating gas bubbles from peripheral tissues during decompression. The main manifestations often involve dysfunction of the central nervous system (CNS) that can lead to permanent disability and long-term residual neurological deficits. Post-decompression shock with plasma volume deficit is a very rare event that has been observed under extreme conditions of high-altitude exposures, experimental diving with helium-oxygen breathing mixtures, and after chronic hyperbaric exposure in compressed-air workers (1–5). To the best of our knowledge, this capillary leak syndrome (CLS) complicating a DCS event in a recreational scuba diver has not been previously reported.

CASE REPORT

A 51-year-old man complained of a sensation of acute vertigo, nausea, and vomiting when climbing on a boat after an uneventful scuba dive with enriched-oxygen breathing mixture (nitrox 25%). The diving parameters were as follows: maximum depth 50 meters sea water (msw), total diving time 60 min, with 30 min of decompression stops from 9 to 3 msw using nitrox 93%. Within 4 days before the incident, he made five deep dives, but did not dive the day before due to an episode of gastroenteritis. The subject was an experienced diver with 1400 logged dives (equivalent to divemaster in Professional Association of Diving Instructors certification) and had no past medical history except for an excess body weight (body mass index 30.5 kg/m^2).

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At presentation in our hyperbaric facility 2 h after the symptoms first appeared and after transportation under first-aid normobaric oxygen, the patient suffered from extreme weakness with unsteadiness, headache, and nausea. Physical examination revealed a spontaneous left beating nystagmus using Frenzel glasses with right-sided asymmetry on a Romberg test, strongly suggesting peripheral vestibular damage. The remainder of the clinical evaluation showed no other pathologic findings, particularly CNS impairment.

Admission laboratory data included a hematocrit of 58% with hemoglobin 20.6 g/dL, total leucocytes count $23 \times 10^3/mm^3$, D-dimer 10,010 ng/mL, serum creatinine 1.6 mg/dL, blood urea 20 mg/dL, and total serum protein 5.7 g/dL. Blood glucose, electrolytes, C-reactive protein, alanine aminotransferase/aspartate aminotransferase, creatine phosphokinase, and prothrombin time were all within normal limits.

In this context of inner ear DCS, the injured diver underwent recompression treatment with hyperbaric oxygen (U.S. Navy table 5) and standard intravenous therapy including acetyl-leucine (vestibular suppressant; 500 mg), metoclopramide (10 mg), methylprednisolone (120 mg), aspirin (250 mg), and normal saline (1 L for 1 h). Preventive anticoagulation (enoxaparin subcutaneous 0.4 mL) was also initiated. He reported slight symptomatic improvement on completion of the hyperbaric treatment and then was transferred to the Emergency Department (ED) for follow-up care and monitoring.

Four hours after admission, the patient complained of abdominal pain and difficulty breathing. Oxygen saturation by oximetry was 91%, and temperature 36.5°C. The pulse was 100 beats/min, and the blood pressure fell to 60/40 mm Hg. Small areas of blue discoloration were noted on the patient's abdomen, which was highly consistent with cutaneous manifestations of DCS, as already observed in previous cases of post-decompression shock (4,5) (Figure 1). He was conscious and oriented without neurological disorder. Cardiac examination revealed no abnormalities. Chest auscultation found bilateral basal crackles without cervical subcutaneous emphysema. Urinary output was low at 100 mL over 4 h.

Repeat laboratory findings confirmed the major leakage of plasma with hypoalbuminemia 18 g/L and the development of acute renal failure (serum creatinine 2.2 mg/dL and blood urea 26 mg/dL). Lipase serum and troponin were normal, and total leukocytes count reached $33 \times 10^3/mm^3$. Arterial blood gas analysis showed the following values: pH 7.31, PaO_2 75 mm Hg, $PaCO_2$ 24.5 mm Hg, HCO_3 12 mmol/L, and lactate 38 mg/dL.

Thoracic and abdominal injected computed tomodensitometry quickly ruled out pulmonary embolism and barotrauma, but revealed generalized edema with pulmonary vascular congestion including pericardial, pleural, and intraperitoneal effusions. Echocardiography confirmed hydropericardium without myocardial dysfunction (left ventricular ejection fraction 75%).

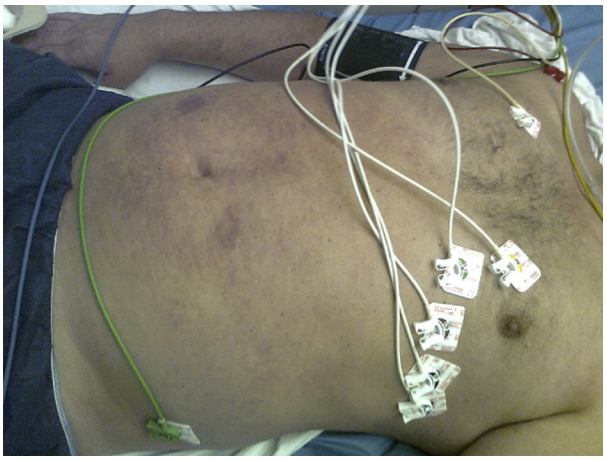

Figure 1. Abdomen covered with purple patches, revealing cutaneous manifestation of decompression sickness accompanying hemodynamic collapse.

In view of his worsening general condition with the developing hypovolemic shock, the patient was transferred to the intensive care unit to apply non-invasive ventilation and to initiate intense albumin 20% infusion.

Over the 2 days following, the clinical signs of anasarca gradually subsided under volume management (400 mL of albumin 20% in 24 h associated with Ringer's lactate solution 3 L/day), normobaric oxygen supply, and low doses of intravenous methylprednisolone (40 mg/day). The patient was transferred to a Medical floor on day 3, where he remained until discharge 7 days later.

Electronystagmography (ENG) with caloric tests during the patient's hospitalization, documented marked right canal paresis (110%), whereas tympanometry and audiometry were normal. Contrast transcranial Doppler ultrasonography failed to reveal the presence of right-to-left shunt. Extensive laboratory testing for possible causes of capillary leak syndrome was carried out. Measurement of serum soluble interleukin (IL)-2 receptor and complement C3a-C4 were negative. A 24-h urinary protein test was below 300 mg. Serum cortisol and thyroid-stimulating hormone were normal. Blood and urine cultures failed to grow pathogenic organisms.

On follow-up 6 weeks after discharge, the patient reported incomplete recovery of inner ear DCS, with transient dizziness triggered by changes in position or head movement that required vestibular rehabilitation. Repeat ENG confirmed residual peripheral vestibular deficit with right-sided caloric hyporeflexia (30%). Serum protein electrophoresis at 1 month was normal, excluding potential underlying monoclonal gammopathy frequently observed in the idiopathic systemic capillary leak syndrome (Clarkson's disease).

DISCUSSION

Vascular bubble formation is the primary pathologic event in the development of decompression sickness. Besides the classic assumption that gas bubbles lead initially to occlusion of capillary blood flow, other mechanisms have been hypothesized by which bubbles may exert secondary deleterious effects. It has been experimentally demonstrated that gas microemboli may interact with vascular endothelium and blood components (platelets and leukocytes, particularly), resulting in activation of the coagulation cascade and host inflammation response leading to thrombi generation, alterations in microcirculation, and hypoxic cell damage similar to pathology seen in ischemia-reperfusion injury (6).

Hemoconcentration, indicated by an increase in hematocrit level, has been frequently observed in experimental DCS using animal models, but the results were less conclusive with human victims of neurological DCS or after uneventful decompression (7–11). Previous clinical work, however, suggests a correlation between the degree of hemoconcentration and the severity of CNS injury in divers with DCS. In a consecutive series of 58 neurological DCS recreational divers, Boussuges and colleagues found that hematocrit values of 48% or greater were significantly associated with the presence of residual deficits at 1 month post-injury (10). It also has been evidenced that neurological outcome, in conjunction with cerebral arterial gas embolism after diving, was strongly associated with hematocrit level at initial admission (12).

Massive plasma deficit with resultant severe hemoconcentration and hypovolemic shock is an uncommon condition affecting divers with a DCS event. The underlying cause has been ascribed to the presence of decompression-induced circulating bubbles that may trigger diffuse endothelial damage by shear stress, resulting in increased vascular permeability with subsequent fluid shift from intravascular compartment into interstitial space (1,3). This hypothesis is strengthened by the beneficial role of plasma expanders in the successful treatment of decompression shock encountered by divers or compressed air workers when prompt recompression remains ineffective (4,5).

The specific mediators contributing to the plasma leakage are unknown, but activation of complement pathway and release of inflammatory cytokines (IL-2, IL-1β) or other factors such as bradykinin, histamine, vascular endothelial growth factor, and lipopolysaccharide endotoxin have been reported to play a role in the increased capillary permeability after endothelial damage (13).

We cannot exclude the possibility that CLS was due not only to the passage of circulating bubbles disrupting the endothelial wall, but also to a combination of vascular gas embolism and exposure to micro-organisms that may exacerbate endothelial dysfunction, as suggested by the episode of gastroenteritis that occurred before the diving accident. Additionally, this gastrointestinal disorder may have contributed to his hypovolemia due to vomiting and diarrhea. On the other hand, close questioning revealed that the symptoms were mild and transient (four episodes of watery diarrhea and one emesis), and laboratory analysis did not exhibit abnormalities consistent with acute sepsis, except the leukocytosis. Actually, the changes in white cell count could be accounted for by the inflammatory and immune activation triggered by the stimulation of endothelial cells and circulating platelets leading to increased expression of leukocyte adhesion molecules (selectins, intercellular adhesion molecules) with concomitant recruitment and activation of leukocytes. Similar findings have been commonly observed in animal DCS models and after scuba diving (14,15).

In the present case, we also found a major and unexpected increase in D-dimer levels, which was interpreted as a severe activation of coagulation. The alteration of this biomarker has been already demonstrated in a previous study involving neurological DCS divers, with a promising role in the prediction of incomplete recovery (16).

CONCLUSION

This report is a reminder that the recreational diver is as susceptible to CLS as the aviator and occupational diver. It is notable that this post-decompression shock was associated with a documented inner ear DCS, whereas previous published cases developed either musculoskeletal pain or transient cerebral manifestations accompanying hemodynamic collapse (3,4). We believe that any injured diver who exhibits elevated hematocrit at presentation to the ED or hyperbaric facility must raise the suspicion for the potential development of CLS, which requires specific treatment using albumin infusion as the primary fluid replacement.

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