Neurogenic Stress Cardiomyopathy Following Aneurysmal Subarachnoid Hemorrhage in a Very Elderly Patient
—Case Report—

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Abstract

A 90-year-old woman presented with aneurysmal subarachnoid hemorrhage (SAH) corresponding to Hunt and Hess grade II. Acute congestive heart failure and pulmonary edema developed following uneventful surgical clipping. Serial electrocardiography and echocardiography led to a diagnosis of neurogenic stress cardiomyopathy (NSC), also known as tako-tsubo cardiomyopathy. The outcome was favorable after supportive therapy with respiratory management and diuretic administration. Neurosurgeons treating SAH must take into account the various general treatment strategy options on a case by case basis after prompt recognition of NSC.

Key words: elderly woman, neurogenic stress cardiomyopathy, pulmonary edema, subarachnoid hemorrhage, tako-tsubo cardiomyopathy

Introduction

Reversible left ventricular dysfunction following aneurysmal subarachnoid hemorrhage (SAH) has been termed neurogenic stunned myocardium, based on the pathological findings. On the other hand, tako-tsubo cardiomyopathy and other terms have also been used for this condition based on the dynamic imaging findings of the left ventricle. Nevertheless, the etiology remains unknown and the terminology is somewhat confusing. Here, we describe such a case in a very elderly patient, which we describe as neurogenic stress cardiomyopathy (NSC) according to the recently proposed terminology. The outcome was favorable despite acute congestive heart failure and pulmonary edema following early surgical intervention.

Case Report

A 90-year-old healthy woman suffered sudden onset of headache and neck pain (day 0, defined by calendar days), and presented to our hospital on day 1. Her consciousness was clear and neurological examination found no abnormalities. Computed tomography (CT) demonstrated diffuse SAH corresponding to Fisher group 3 (Fig. 1A), whereas subsequent CT angiography disclosed a left vertebral artery-posterior inferior cerebellar artery (VA-
Fig. 1 Computed tomography (CT) scan (A) demonstrating diffuse subarachnoid hemorrhage. CT angiogram (B) revealing a vertebral artery-posterior inferior cerebellar artery aneurysm.

Fig. 2 Electrocardiograms, on day 1 (A) showing ST segment elevations in leads V1 to V4 and a negative T wave in lead V4, and on day 2 (B) showing deep inverted T-waves in leads V2 to V6 with a prolonged Q-T interval.

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PICA) aneurysm (Fig. 1B). Electrocardiography (ECG) showed ST segment elevations in leads V1 to V4, inverted T wave in lead V4 (Fig. 2A), and sinus rhythm with a heart rate of 85 beats per minute. She did not complain of chest pain or dyspnea. Chest radiography showed no abnormalities. Laboratory examinations revealed mildly elevated levels of cardiac enzymes, with creatine kinase of 295 IU/l, aspartate aminotransferase of 40 IU/l, white blood cell count of 12 430/l, and C-reactive protein of 2.2 mg/dl.

Surgical clipping was performed on day 2. Before induction of anesthesia, ECG revealed significant changes, including deep inverted T-waves in leads V2 to V6 with a prolonged Q-T interval (Fig. 2B). Administration of dopamine was required to maintain blood pressure during anesthesia. The VA-PICA aneurysm was successfully obliterated. However, after recovery from anesthesia, the patient awoke with agitation and suddenly developed transient ventricular tachycardia concomitant with hypotension. Shortly thereafter, oxygenation deteriorated, which required positive end-expiratory pressure support. Postoperative chest radiography revealed pulmonary edema and blood pressure was stabilized by short duration dopamine support. The patient was then treated with administration of furosemide under ventilatory support.

A cardiologist was consulted on day 3 and a diagnosis of acute congestive heart failure due to tako-tsubo cardiomyopathy was made based on the echocardiographical findings. Echocardiography on day 6 revealed severe hypokinesis in the mid-anterior and apical regions of the left ventricle (Fig. 3A, B). The ejection fraction was 45%. The level of cardiac troponin T was 0.09 ng/ml (normal <0.1 ng/ml, obtained on day 6), and brain natriuretic peptide had increased to 1690 pg/ml (normal range 0 to 18.4 pg/ml, obtained on day 3). The patient was extubated on day 8.

Coronary angiography, left ventriculography, and cerebral angiography were performed on day 19. Coronary angiography showed normal coronary arteries, whereas left ventriculography revealed mild hypokinesis of the apex. The ejection fraction was 72%. Cerebral angiography showed complete obliteration of the VA-PICA aneurysm and no remarkable vasospasm. Normalization of left ventricular dysfunction was confirmed by follow-up echocardiography (Fig. 3C, D). The patient received a ventriculoperitoneal shunt for hydrocephalus and was dis-
charged without neurological deficits. Follow-up ECG at 3 months showed the inverted T waves had returned to nearly normal.

Discussion

Myocardial stunning mimicking acute myocardial infarction was first reported in Japan and has also been described as tako-tsubo cardiomyopathy.\(^\text{15}\) The condition is typically observed as transient hypokinesia of the apical region, in contrast to hypokinesia of the basal walls.\(^\text{15}\) Similar etiology was pointed out following aneurysmal SAH\(^\text{9}\) and has been increasingly recognized under various terms, such as neurogenic stunned myocardium and reversible neurogenic left ventricular dysfunction.\(^\text{2,7}\)

However, whether these conditions have substantially the same pathogenesis remains unknown, though catecholamine surge is considered to be important. Recently, the new term ‘NSC after SAH’ was proposed (Table 1).\(^\text{24}\) Early ST segment elevation in ECG concomitant with mildly elevated cardiac enzymes can be observed in the acute phase in affected patients. In addition, inverted T waves are commonly seen within 24 hours, as in the present case. Importantly, coronary artery angiogram findings are normal despite these observations mimicking acute myocardial infarction. In these cases, regional wall motion abnormalities with reduced cardiac EF (≤ 40%) are observed as global left ventricular wall dysfunction, after which pulmonary edema can develop. Eight cases associated with poor SAH corresponding to Hunt and Hess grades III and IV, pulmonary edema requiring prolonged intubation, and cerebral vasospasms have been reported.\(^\text{23}\) Hypothalamic dysfunction, exaggerated sympathetic activity, and catecholamine surge are considered to be the main pathogenic factors of NSC after SAH.

We identified 29 reports including a total of 57 cases written in English up to January 2011 in searches of PubMed using various terms, such as ‘tako-tsubo cardiomyopathy AND subarachnoid hemorrhage’ and ‘neurogenic stunned myocardium AND subarachnoid hemorrhage’.\(^\text{1,4,6–9,11–14,16,17–23,25,26,29,32–39}\) Distribution by age and sex (excluding our case) revealed a clear predominance in females (86%), with ages ranging from 28 to 83 years (mean 56 years) (Fig. 4). The present patient was older than all others previously reported.

Cardiac prognosis related to this specific left ventricular dysfunction is favorable, because of the self-limiting nature within a few weeks.\(^\text{2,3}\) However, NSC is challenging to treat in patients with poor grade SAH complicated by cerebral vasospasms. Furthermore, NSC leading to cardiopulmonary arrest,\(^\text{36}\) cardiogenic shock requiring an intra-aortic balloon pump,\(^\text{14,20,21}\) and sudden death\(^\text{30}\) have been reported. Prompt recognition, consultation with a cardiologist, and intensive cerebral and cardiovascular monitoring are important, although no specific treatments have been established.\(^\text{21}\) To differentiate NSC from acute myocardial infarction, measurement of cardiac troponin (cTn), including its 2 isoforms cTn T and cTn I, would be helpful.\(^\text{7}\) The peak value of cTn observed within 3 days after onset of SAH in NSC is much less than that seen in myocardial infarction. However, cTn elevation was observed in 20% to 40% of SAH patients with and without NSC.\(^\text{23,28,31}\) Routine echocardiography would be helpful to detect NSC.

The inotropic agents dobutamine and milrinone can be used to improve cerebral hemodynamics.\(^\text{5,27}\) In addition, the β-blockers propranolol and atenolol may prevent catecholamine-induced myocardial damage.\(^\text{10}\) Triple H therapy can cause fluid overload, and induce or worsen NSC with the use of the exogenous catecholamines dobutamine and norepinephrine.\(^\text{37}\) If vascular resistance and blood pressure are maintained, the non-catecholamine agent milrinone may be promising to treat both cerebral vasospasm and NSC.\(^\text{5,27}\) Neurosurgeons may encounter a dilemma when planning to obliterate a ruptured cerebral aneurysm, regarding whether to employ an early or delayed strategy with surgical clipping or an endovascular procedure. Although the choice can be delayed until the respiratory and hemodynamic states are stabilized, dependent on both SAH grade and NSC,\(^\text{14,16}\) but the risk of re-rupture is inevitable.\(^\text{16}\) Favorable outcomes of surgery for good grade SAH in elderly patients aged 70–89 years have been reported,\(^\text{18}\) but endovascular surgery techniques have increased in importance.

The present case of NSC was atypical in regard to good grade SAH and very high age, although early surgical

### Table 1 Clinical characteristics of cases of neurogenic stress cardiomyopathy\(^\text{24}\)

<table>
<thead>
<tr>
<th>Cardiac enzyme</th>
<th>modest elevation</th>
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<tbody>
<tr>
<td>Electrocardiogram</td>
<td>ST elevation, T-wave inversion, QT prolongation</td>
</tr>
<tr>
<td>Echocardiogram</td>
<td>left ventricular RWMA beyond single vascular territory</td>
</tr>
<tr>
<td>Coronary angiogram</td>
<td>normal</td>
</tr>
<tr>
<td>Chest radiogram</td>
<td>pulmonary edema</td>
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RWMA: regional wall motion abnormalities.

Fig. 4 Bar graph showing distribution by age and sex of reported neurogenic stress cardiomyopathy cases. Open columns: female, closed columns: male.
treatment was successful. Additional cases are likely to be treated in the future with an early endovascular procedure. Neurosurgeons treating SAH must take into account treated in the future with an early endovascular procedure.

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