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CASE REPORT

A case of neurogenic myocardial stunning presenting transient left ventricular mid-portion ballooning simulating atypical takotsubo cardiomyopathy

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KEYWORDS
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Summary A 57-year-old female patient, who was initially suspected to have subarachnoid hemorrhage, was admitted to our hospital. She experienced severe dyspnea and chest pain owing to pneumonia on the fourth admission day. Electrocardiography showed ST-segment elevation in leads V_2 through V_5, and echocardiography revealed hypokinetic left ventricular wall motion. No stenosis was found in the coronary arteries by urgent coronary angiography. However, left ventriculography revealed that the basal and apical areas were hyperkinetic and the mid portion was akinetic. After a month, left ventricular wall motion was improved and coronary artery spasm provocation tests were negative. Although the clinical course of this patient was similar to that of neurogenic myocardial stunning, the shape of her left ventricle was not typical.

Introduction

Neurogenic myocardial stunning and takotsubo cardiomyopathy are myocardial disorders characterized by acute myocardial infarction-like electrocardiography (ECG) changes and transient regional hypokinesis of the left ventricular wall. In this condition, left ventricular wall dyskinesis is not consistent with the coronary artery distribution, and hypokinesis is limited to the apical region [1—3]. This report presents a case of a probable variation of neurogenic myocardial stunning, which showed a clinical course similar to that of takotsubo cardiomyopathy but hyperkinesis in the basal

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and apical areas and akinesis in the middle of the left ventricle.

Case report

A 57-year-old female underwent emergency examination at the Department of Neurosurgery of our hospital with a primary symptom of headache in June 2005. As head computed tomography suggested subarachnoid hemorrhage (Fig. 1), the patient was admitted on the same day. The next day, coil embolization and ventricular drainage were performed under general anesthesia. The subsequent course was uneventful, but hypoxia, probably due to aspiration pneumonia, occurred on the 4th hospital day, and artificial ventilation was performed again. After re-intubation, ECG showed marked ST-segment elevation in leads V_{2-5} compared with the trace on admission (Fig. 2A and B), and echocardiography revealed circumferential hypokinesis in the middle of the left ventricle

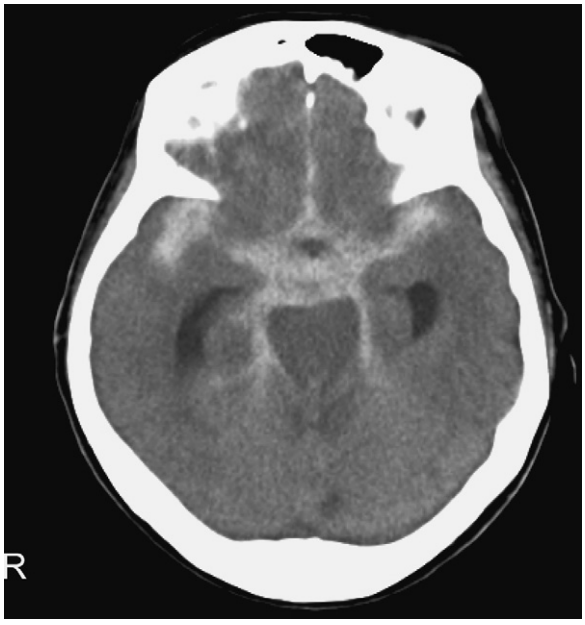

Figure 1 Computed tomography scan of the head.

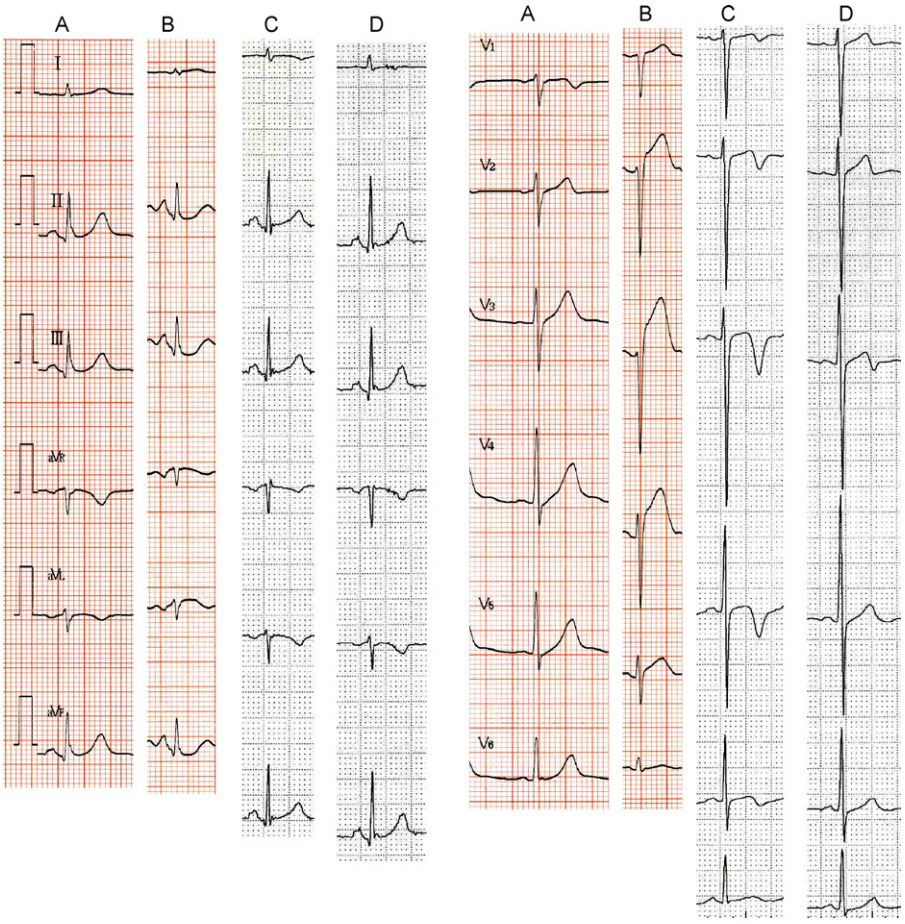

Figure 2 Time course of electrocardiography. (A) Electrocardiography on admission. (B) Electrocardiography at onset showing ST elevation in V_{2-5}. (C) Electrocardiography 7 days after the onset showing ST improvement and negative T. (D) Electrocardiography 2 months after the onset showing normalization, except for negative T wave in V_3.

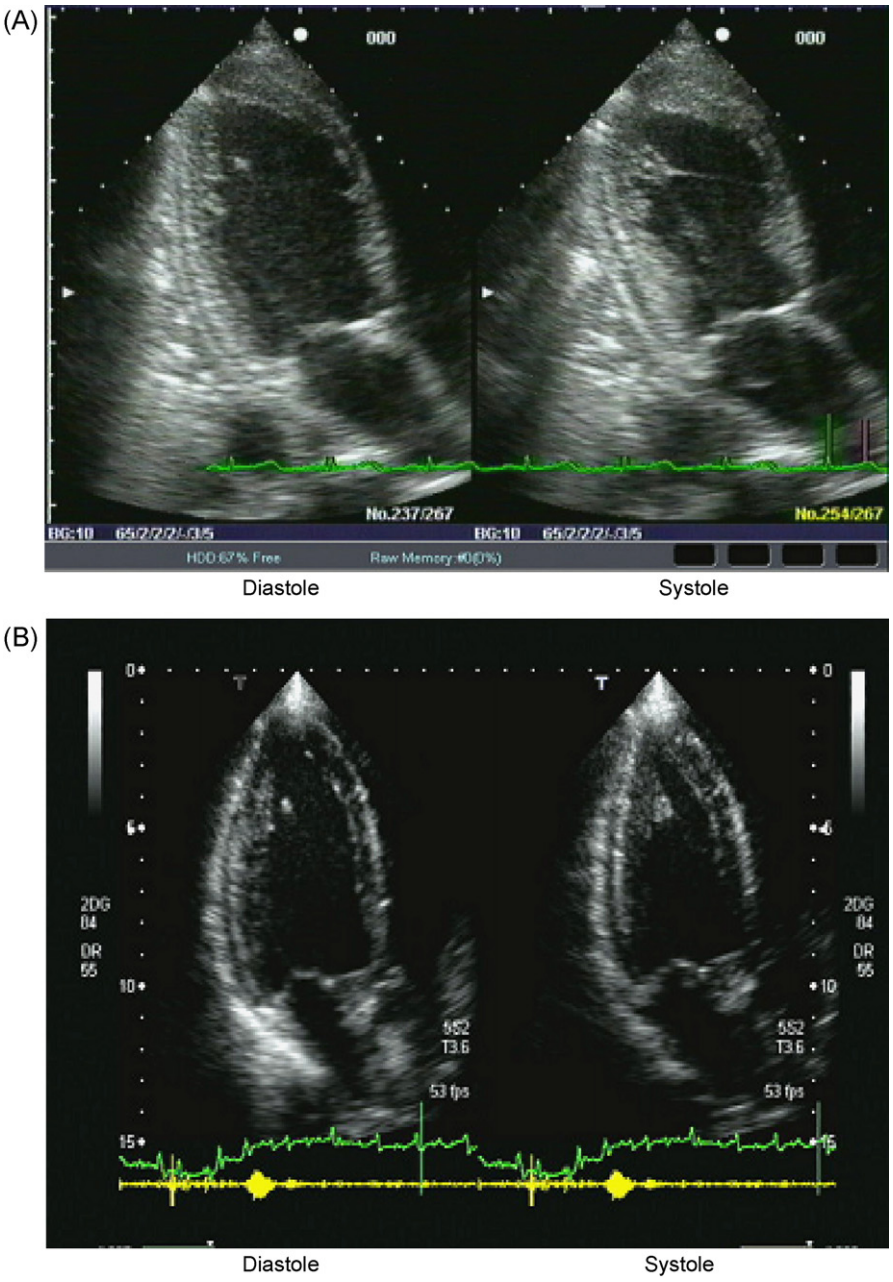

Figure 3 Echocardiography. (A) In the acute phase, left ventricular ballooning was observed in the mid portion as shown by ventriculography. The basal and the apical areas were hyperkinetic (end-diastolic dimension: 50 mm, end-systolic dimension: 42 mm, ejection fraction: 32%). (B) After 2 months, the left ventricular dysfunction had almost recovered (end-diastolic dimension: 38 mm, end-systolic dimension: 24 mm, ejection fraction: 56%).

(Fig. 3A). In consideration of the possibility of acute myocardial infarction, emergency cardiac catheterization was performed. Coronary angiography showed no significant stenosis in the left or right coronary artery (Fig. 4). Left ventriculography revealed akinesis in the middle of the left ventricle and hyperkinesis of the basal and apical regions (Fig. 5A). The maximum creatinine kinase level was not increased, at 179 IU/l, but the brain natriuretic peptide level was 1475 pg/ml, and the plasma noradrenaline level was 1304 pg/ml, at the onset, showing marked increases (Fig. 6). Conservative treatment for heart failure was performed, followed by an uneventful course. ECG on the 7th hospital day showed alleviation of ST-segment elevation noted in precordial leads and the negative conversion of T-waves (Fig. 2C), and these T-waves normalized on the 60th hospital day (Fig. 2D). When cardiac catheterization was performed again after about a month, the wall motion

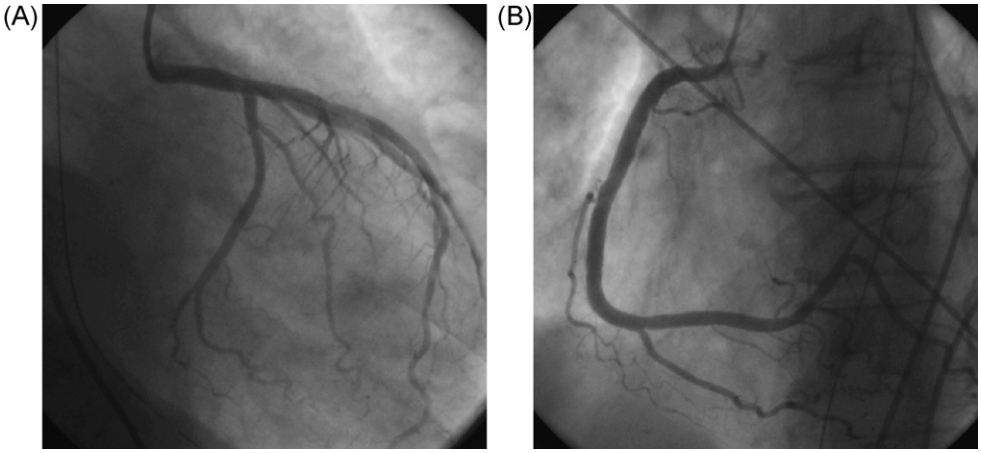

Figure 4 Coronary angiography. Coronary angiography showed normal left coronary artery (A) and normal right coronary artery (B).

abnormality in the middle of the left ventricle observed in the acute period by left ventriculography was reduced (Fig. 5B). The coronary spasm provocation test using acetylcholine under coronary angiography was performed simultaneously, but no spasm was induced (Fig. 7). On echocardiography at discharge, the left ventricular wall motion had nearly normalized, and the left ventricular ejection fraction was 56%. No decrease in wall thickness or increase in echo signals such as those observed in ischemic heart disease was noted (Fig. 3B). The patient was transferred to another

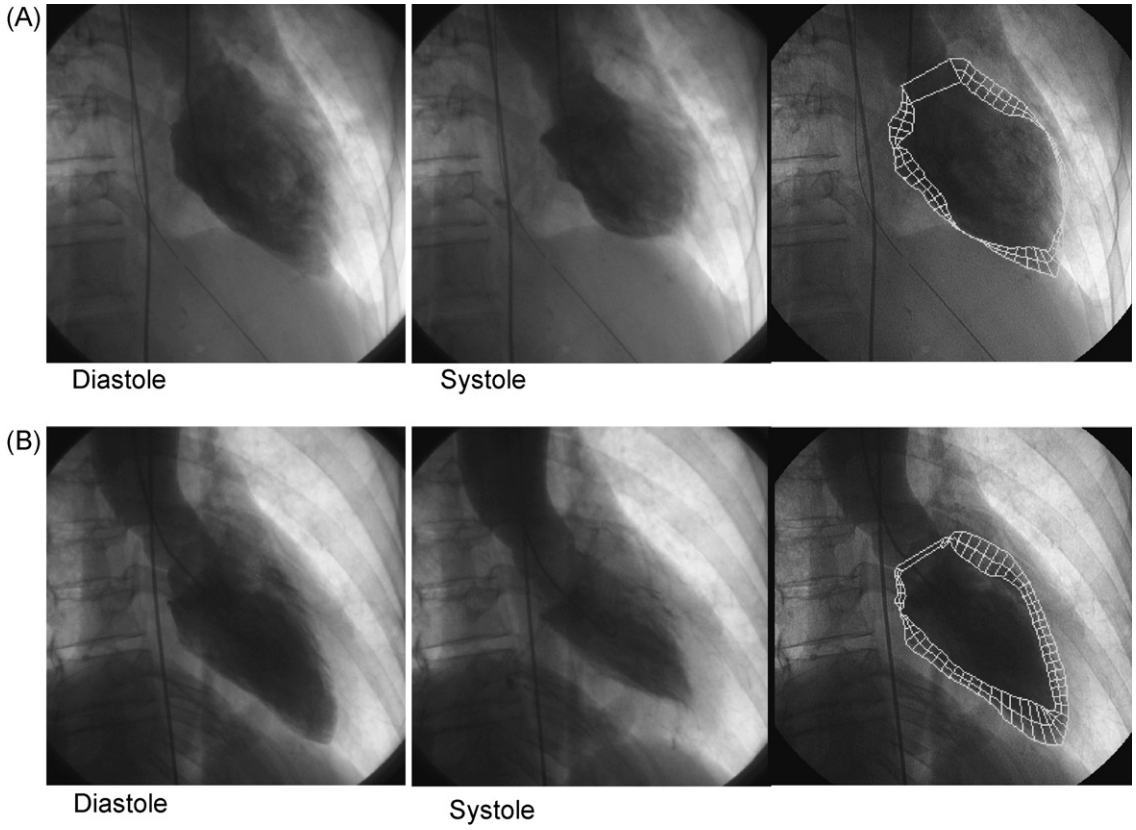

Figure 5 Left ventriculography. (A) In the acute phase, left ventriculography revealed that the basal and the apical areas were hyperkinetic and the mid portion was akinetic. (B) After a month, left ventriculography revealed almost normal contraction.

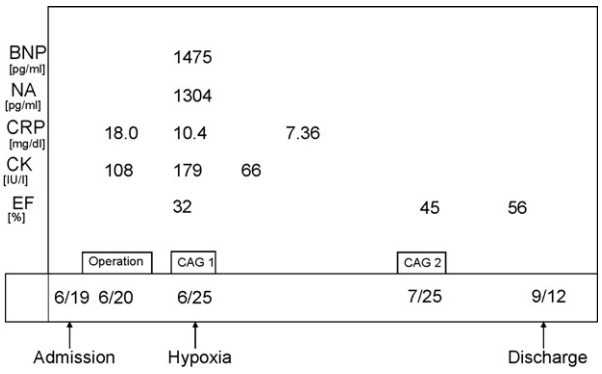

Figure 6 Clinical course. The CK level reached 179 IU/l at onset and decreased. Contraction of the left ventricle improved gradually. BNP, brain natriuretic peptide; NA, noradrenaline; CRP, C-reactive protein; CK, creatine kinase; EF, ejection fraction; CAG, coronary angiography.

hospital for neurosurgical rehabilitation in September 2005.

Discussion

Although the clinical course of this patient suggested neurogenic myocardial stunning, the pattern of left ventricular dyskinesis, i.e., akinesis in the middle of the left ventricle and hyperkinesis in the basal and apical regions, differed from that in typical cases. Systolic morphological abnormalities such as the inversed takotsubo type showing akinesis in the basal region and hyperkinesis in the apical region [4] (chestnut type [5]) and the sandglass type showing akinesis in the apical and basal regions and hyperkinesis in the middle of the left ventricle [6] have been reported sporadically as variations of takotsubo cardiomyopathy.

The mechanism of the wall motion abnormalities observed in this patient could not be determined. Multi-vessel coronary spasms and an excess of catecholamines have been suggested as possible causes of takotsubo cardiomyopathy [7]. In this patient, no significant stenosis of the coronary artery was noted, and no spasm was induced in the coronary spasm provocation test using acetylcholine. In addition, akinesis of the middle of the left ventricle could not be explained by the distribution of the coronary artery. However, coronary vasospasm was often unable to be induced by acethylcholine or ergometrine after acute myocardial ischemia. These results may not be always contradictory to multi-vessel coronary spasms.

Concerning catecholamines, a state of Ca-overloading in cells associated with increased cell membrane permeability and the involvement of free radicals and oxidative stress have been suggested as mechanisms of myocardial disorders [8]. While the plasma catecholamine concentration was abnormally high in this patient immediately after the onset, the density of sympathetic nerve terminals has been anatomically reported to be high in the basal region and low in the apical region [9], and myocardial disorders due to catecholamines alone do not explain the wall motion abnormalities in this patient. There may be individual variation in the sympathetic innervation of the left ventricle, but evaluation of this point was impossible, because we did not perform cardiac radionuclide examinations in this patient.

A similar hypothesis was discussed in a previous case report of pheochromocytoma [10]. In this patient pheochromocytoma was ruled out due to the symptom and abdominal ultrasound examination.

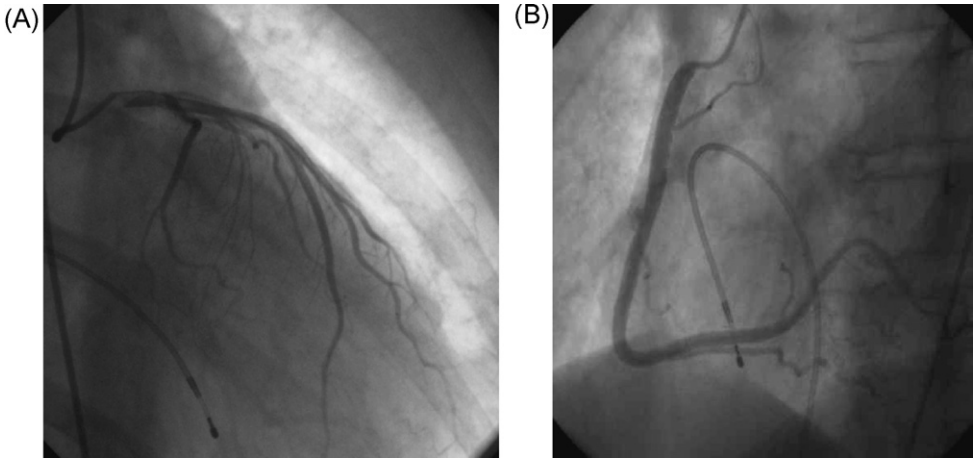

Figure 7 Spasm provocation test. Spasm provocation test did not show the severe vasoconstriction in the left coronary artery (A) and right coronary artery (B).

In the acute period of subarachnoid hemorrhage, changes in ECG and clinical symptoms resembling those of ischemic heart disease including myocardial infarction are often observed [11—13]. A theory concerning the mechanism of these changes that is widely supported is as follows: Subarachnoid hemorrhage causes hemorrhage or ischemia of the hypothalamus or its compression due to brain edema, and, as the hypothalamus is damaged, a large amount of endogenous catecholamines is released and causes extensive spasms of systemic peripheral blood vessels including the coronary artery, resulting in ECG changes and clinical symptoms resembling those of ischemic heart disease [14—16]. In this patient, atypical neurogenic myocardial stunning after subarachnoid hemorrhage was suspected. However, atypical takotsubo cardiomyopathy owing to physical or mental stress might be related to this case.

The mechanism of such myocardial disorders must be clarified by comparatively analyzing cases showing various types of wall motion abnormalities.

This report presented a case of atypical neurogenic myocardial stunning presenting hyperkinesis of the basal and apical regions and akinesis in the middle of the left ventricle.

References

[1] Dote K, Sato H, Tateishi H, Uchida T, Ishihara M. Myocardial stunning due to simultaneous multivessel coronary spasms: a review of 5 cases. J Cardiol 1991;21:203—14.
[2] Kawai S, Suzuki H, Yamaguchi H, Tanaka K, Sawada H, Aizawa T, et al. Ampulla cardiomyopathy ('Takotusbo' cardiomyopathy)—reversible left ventricular dysfunction: with ST segment elevation. Jpn Circ J 2000;64:156—9.
[3] Nyui N, Yamanaka O, Nakayama R, Sawano M, Kawai S. 'Tako-Tsubo' transient ventricular dysfunction: a case report. Jpn Circ J 2000;64:715—9.
[4] Takeno Y, Eno S, Hondo T, Matsuda K, Zushi N. Pheochromocytoma with reversal of tako-tsubo-like transient left ventricular dysfunction: a case report. J Cardiol 2004;43:281—7.
[5] Oguri A, Uozumi H, Sawaki D, Kim M, Kobayakawa N, Fukushima K, et al. ''Chestnut-shaped'' transient regional left ventricular hypokinesis with abnormal myocardial fatty acid metabolism, not corresponding to the coronary artery territories: a case report. J Cardiol 2004;43:273—80.
[6] Sugi Y, Muro A, Yoshida T, Takajyo Y, Matsuoka H, Ohuchida M, et al. Pheochromocytoma presenting as acute heart failure. Nippon Naika Gakkai Zasshi (J Jpn Soc Intern Med) 2002;91:467—9.
[7] Kawai S. Ampulla-shaped ventricular dysfunction or ampulla cardiomyopathy? Kotojun 2000;48:1237—48.
[8] Drislane FW, Samuels MA, Kozakewich H, Schoen FJ, Strunk RC. Myocardial contraction band lesions in patients with fatal asthma: possible neurocardiologic mechanisms. Am Rev Respir Dis 1987;135:498—501.
[9] Randall WC, Kaye MP, Hageman GR, Jacobs HK, Euler DE, Wehrmacher W. Cardiac dysrhythmias in the conscious dog after surgically induced autonomic imbalance. Am J Cardiol 1976;38:178—83.
[10] Yamanaka O, Yasumasa F, Nakamura T, Ohno A, Endo Y, Yoshimi K, et al. 'Myocardial stunning'-like phenomenon during a crisis of pheochromocytoma. Jpn Circ J 1994;58:737—42.
[11] Yamaguchi T, Shimizu Y, Ono N, Unno M, Nishikawa H, Kakuta Y, et al. A case of subarachnoid hemorrhage with electrocardiographic and echocardiographic changes simulating transmural myocardial infarction. Jpn J Med 1991;30:142—5.
[12] Sakamoto H, Nishimura H, Imataka K, Ieki K, Horie T, Fujii J. Abnormal Q wave, ST-segment elevation, T-wave inversion, and widespread focal myocytolysis associated with subarachnoid hemorrhage. Jpn Circ J 1996;60:254—7.
[13] Rorbakken G, Brunvand H, Gundersen T, Farstad G. Cardiopulmonary complications in acute subarachnoid hemorrhage. Tidsskr Nor Laegeforen 1998;118:3430—4.
[14] Sakr YL, Ghosn I, Vincent JL. Cardiac manifestations after subarachnoid hemorrhage: a systematic review of the literature. Prog Cardiovasc Dis 2002;45:67—80.
[15] Lanzino G, Kongable GL, Kassell NF. Electrocardiographic abnormalities after nontraumatic subarachnoid hemorrhage. J Neurosurg Anesthesiol 1994;6:156—62.
[16] Zaroff JG, Rordorf GA, Titus JS, Newell JB, Nowak NJ, Torchiana DF, et al. Regional myocardial perfusion after experimental subarachnoid hemorrhage. Stroke 2000;31:1136—43.