Acute spontaneous spinal subdural hematoma presenting with Takotsubo cardiomyopathy: a rare case report and literature review

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- review
- treatment
- case report

Introduction
Spontaneous spinal subdural hematoma (SSDH) is characterized by the sudden onset of back pain or headache and can lead to severe consequences (1). The most common causative factors are anticoagulant therapy and dysfunction of blood coagulation, followed by atrogenic procedures using a needle. The mechanisms of SSDH development are still unclear, as the subdural cavity of the spinal cord lacks a bridging vein. In patients with SSDH, the duration from the onset of hemorrhage to clinical presentation varies from a few hours to a few months (2). Moreover, most patients with SSDH present with sudden back pain followed by sensorimotor changes, neurogenic bladder, and autonomic dysfunction. While the prognosis of SSDH is determined by the severity of neurologic deficits, the influence of early cardiovascular changes secondary to SSDH cannot be ignored. Herein, we reported a case of a patient with acute SSDH and presenting with Takotsubo cardiomyopathy (TTC) and reviewed the relevant literature.

Case report
Patient Information
A 52-year-old woman in good health developed tingling of the thoracic back with associated numbness and weakness in the bilateral lower limbs while taking a bath 1 day ago. The thoracic back tingling was unfixed, and the patient was unable to walk and had an obvious sense of zonesthesia below the T6 level. She denied bladder dysfunction, chest pain, or distress. She also had no hypertension, pulmonary, gastrointestinal, coagulation disorders, cardiovascular problems, urinary-tract disorder, trauma, or invasive therapy. The patient was taken to the emergency department immediately after developing symptoms.
Physical examination

The vital signs on arrival were temperature (T) 37.7°C, pulse (P) 94 beats/min, respiration rate (RR) 18 breaths/min, oxygen saturation (O₂ sat): 100%, and blood pressure (BP) 102/56 mmHg. Neurological examination revealed hypoesthesia below T6 and normal lower-limb muscle strength (Medical Research Council (MRC) grade 5/5). On admission, the vital signs were T 37.1°C, P 74 beats/min, RR 18 breaths/min, O₂ sat: 100%, and BP 147/74 mmHg. Physical examination revealed a sensory deficit below the level of T6, bilateral extensor plantar reflexes, paraparesis (MRC grade 4/5 bilaterally in the distal and proximal leg musculature), loss of perineal sensation, negative Hoffman signs, and positive Babinski signs.

Timeline

Six hours after admission, the patient was catheterized because of urinary retention. Nine hours later, the patient experienced an aggravation of pain, profuse sweating, and tachycardia (heart rate was up to 163 beats/min). Self-inflicted scratches were seen on her chest. She had normal bladder sensation but difficulty urinating, progressive loss of muscle strength, and sinus tachycardia. Urethral catheterization was performed for difficult urinating. To treat the pain and stress caused by the sinus tachycardia or trauma, she was given dezocine for pain and esmolol to control the heart rate. One hour later, her O₂ sat dropped to 82% and BP decreased. Blood gas analysis showed a lactic acid concentration of 6.7 mmol/L, indicating microcirculation hypoxia. Considering the patient had heart failure, furosemide was given for diuretic therapy.

She was then transferred to the intensive care unit (ICU) for further treatment. The detailed timeline is shown in Fig. 1.

Laboratory examinations

Laboratory evaluation showed leukocytosis (30 600 cells/µL) (neutrophils 90.1%), an increased brain natriuretic peptide (BNP) concentration of 320 pg/mL, increased cardiac troponin I (cTNI) concentration of 8.97 ng/mL, and normal coagulation screening test results.

Imaging examinations

MRI revealed a subdural hematoma from C7-T7 anterior to the spinal cord (Fig. 2A and F) and ECG showed sinus tachycardia (Fig. 3A). Echocardiography showed left ventricular ejection fraction (LVEF) of 28%. The contrast-enhanced MRI revealed no spinal arteriovenous malformations (Fig. 2E and J).

Final diagnosis

We organized a multi-disciplinary team (MDT) meeting with doctors from the departments of Thoracic Surgery, Vascular Surgery, Cardiology, ICU, Respiratory Medicine, and Orthopedic. Based on the clinical findings and physical examination findings, the patient was diagnosed with acute spontaneous spinal subdural hematoma presenting with TTC.

Therapeutic intervention

Because of the extensive craniocaudal extension of SSDH, conservative treatment was chosen, with readiness for surgery in case of progressive worsening. The patient was...
SSDH with Takotsubo cardiomyopathy

Treated with dopamine, spironolactone, and furosemide for heart failure, metoprolol for heart rate control, and methylprednisolone as pulse therapy.

Follow-up and outcomes

Fortunately, these symptoms and signs were alleviated after the administration of heart failure medications and other supportive therapies, and the vital signs were stable (O₂ saturation: 95–100%, BP: 95–100/60–80 mmHg, P: 77–100 beats/min). Significant recovery was shown on repeat MRI (decreased area of SSDH, Fig. 2C and H), ECG (reduced heart rate, Fig. 3B), and echocardiography (LVEF: 42%, Fig. 3D) together with normal cTNI (Fig. 3E), and BNP (Fig. 3F) concentrations.

Figure 2
MRI performed with T2-weighted sagittal and axial views. There are subdural hematomas (white arrow) from C7-T7 anterior to the spinal cord on T2-weighted images during hyperacute phase (A and F) and acute phase (B and G). Repeat MRI shows marked reduction in the area of subdural hematomas (white arrow) after 25 days (C and H). There are adhesions in the subdural space (black arrow) in repeated MRI after 3 months (D and I). Contrast-enhanced MR revealed no spinal arteriovenous malformations (E and J).

Figure 3
Operational changes in ECG and echocardiography, and cTNI and BNP concentrations during hospitalization. The ECG shows frequent sinus tachycardia (A). These abnormalities returned to normal 2 days later (B). The left ventricle was slightly enlarged with global hypokinesis, LVEF 28% (C). Ten days later, the motion of the LVEF was 42% (D). The concentrations of cTNI (E) and BNP (F) were markedly increased at the beginning of heart failure but returned to normal ranges after 2 weeks. BNP, brain natriuretic peptide; cTNI, cardiac troponin I; LVEF, left ventricular ejection fraction.
At 28 days after treatment, the patient felt numbness on the outer side of both calves and the zonesthesia had disappeared. At the time of discharge, she still had involuntary painful twitching in the back and abdomen. However, the area of pain was reduced, the duration of pain was shorter, and the severity of pain was much less than at admission. Physical examination revealed that the muscle strength was MRC grade 4 in the right distal and proximal leg musculature and MRC grade 5 in the left, with tactile pain on the left side that was lower than the right side; the patient had positive ankle clonus and Babinski signs, hyperactive knee tendon reflexes and ankle reflexes, and no patellar clonus bilaterally. A repeat MRI performed 3 months after the patient was discharged showed adhesions in the subdural space (Fig. 2D and I). During a telephone interview at 11 months, the patient reported sometimes feeling right calf numbness with bilateral lower extremity strength of MRC grade 5 and was very satisfied with the results of our treatment.

Discussion

TTC, also known as stress cardiomyopathy, is a syndrome characterized by transient local systolic dysfunction of the left ventricle that often manifests as chest pain, dyspnea, ECG changes, and myocardial biomarker release, similar to acute coronary syndrome. TTC was first described by Sato et al. in 1990 (3). It is mainly caused by mental or physical stress and occurs relatively frequently in postmenopausal women. It is usually a self-limited cardiomyopathy, with left ventricular dysfunction recovering within 1 month. However, some cases result in serious complications or death (4). The pathogenesis of TTC is still unclear; however, sympathetic hyperactivity and massive release of catecholamines are currently recognized as the main mechanisms. Excessive catecholamines directly damage cardiomyocytes or indirectly cause transient microvascular dysfunction and ischemic injury (5). It has been reported that normal ventricular contraction is gradually restored after the administration of adrenergic receptor blockers in a rat emotional stress-induced model of TTC (6).

Matos-Souza et al. reported that subjects with spinal cord injury (SCI) have impaired LVEF diastolic function in comparison with subjects without SCI (7). And there have multiple cases indicating that the cardiac dysfunction is related to SCI (8, 9, 10). Once SSDH causes SCI, clinicians should consider the possibility of cardiovascular system dysfunction. The activities of the heart and other internal organs are regulated by descending pathways of sympathetic neurons located in the mid-lateral column of the spinal cord from T1 to L2. Therefore, patients with high SCI have a higher incidence and adjusted hazard ratio of cardiometabolic morbidities (11). As the impaired sympathetic nerve that innervates the heart and blood vessels, leading to the relative advantage of vagus nerve function, which revealed deficiency in control of the autonomic nervous system (12).

In our case, a middle-aged woman experienced tingling of the thoracic back without determined reasons. The MRI revealed subdural hematoma from C7-T7 and the laboratory examination showed elevated BNP and cTNI levels. Following the development of neurological symptoms, her BP decreased and she developed sinus tachycardia and rapidly developed acute heart failure. The cardiac symptoms resolved after treatment with cardiotonic, diuretic, and vasodilator medications. The patient was a postmenopausal woman with no prior risk factors for cardiovascular disease. After supportive therapies, repeat measurements of cTNI and BNP and repeat echocardiography revealed significant recovery. This shows that the myocardial injury is transient and reversible, ruling out myocarditis and acute myocardial infarction. Besides, the patient did not indicate that she experienced any intense stressors before her condition developed. Therefore, we deduced that the subdural hematoma leads to SCI, which results in the occurrence of TTC. Previous studies have shown that TTC can occur secondarily to neurological diseases, such as subarachnoid hemorrhage, stroke, and seizures (4). Nevertheless, TTC caused by SSDH is extremely rare.

We have searched the EMBASE, PubMed, CNKI, and Web of Science databases using the terms ‘spinal subdural hematoma and (heart or Takotsubo)’ and identified only two reported cases of acute heart failure induced by SSDH before March 2022 (13, 14). The clinical profiles of these cases, including publication year, patient’s age/sex, hematoma location, cause of bleeding, elevation of cTNI and BNP, treatment, follow-up duration, and outcome were summarized in Table 1. Including our case, a total of three patients have been reported to have TTC and SSDH, comprising two women and one man with an average age of 35 years. Unlike previously reported cases, our case involved a postmenopausal woman with an earlier onset time and a more extensive range of SSDH. The early increase in BNP was not obvious, and the primary manifestation was a sharp increase in cTNI. The two previously reported cases both involved young people who did not undergo repeat MRI and mainly showed a significant increase in BNP in the early stage, without an obvious increase in troponin. All of these three cases recovered well after conservative treatment. These two previous patients were not followed up after being discharged. Among patients with SSDH, conservative treatment may be a feasible option for those with spontaneous resolution of symptoms or only mild transient neurological deficits, while emergency surgery may be more beneficial for patients with paralysis or progressive reductions in muscle strength (2, 15). Conservative treatment is even effective...
for patients with severe paraparesis after spontaneous acute SSDH (16). Chong et al. reported a case of a patient with acute SCI due to a lateral perimedullary arteriovenous malformation which presented with TTC and recovered uneventfully after urgent surgery (17). After the initial SSDH diagnosis, we also considered urgent surgery for our patient. However, because of the extensive range of SSDH and the acute cardiac insufficiency, the MDT meeting consensus was to perform a conservative treatment. All three reported patients with TTC and SSDH recovered and were discharged after conservative treatment. The patient in our case recovered well after 11 months of follow-up. The adhesions in the subdural space shown on repeat MRI performed 3 months after the patient was discharged were thought to be caused by scar tissue that formed after hematoma resorption.

Our case might have clinical implications. First, we confirmed previous reports that conservative treatment is effective for TTC secondary to SSDH. Secondly, we have strong evidence that our patient’s SSDH was spontaneous. SSDH is a rare disease, and the etiology is often related to trauma, abnormal coagulation function, or arteriovenous malformation (18, 19). In our case, Contrast-Enhanced MRI ruled out arteriovenous malformation, and the patient denied trauma and had no obvious abnormal coagulation function; therefore, we considered the SSDH to be spontaneous. Thirdly, our patient had no complications during a relatively long follow-up period. During the entire 11-month period, the patient had no recurrence of cardiac complications, and the repeat MRI showed that the SSDH led to adhesions in the subdural space after hematoma absorption.

Our study has some limitations. First, at the onset of tachycardia, our cardiologist did not recognize that the patient had TTC. The tachycardia was initially misdiagnosed as sinus tachycardia and treated with a heart-rate-lowering medication, leading to the treatment delay. Secondly, the mechanism of TTC secondary to SSDH is still unclear. Thirdly, we did not assess the concentration of circulating catecholamines, and the relationship between TTC and the concentration of catecholamines is unclear. Fourthly, coronary angiography and ventricular angiography were not performed because the patient was in a critical condition. Fifthly, only two other cases were included in our review; more clinical studies should be conducted in the future.

### Conclusion

This rare case broadens the symptom spectrum of SSDH and suggests that initial conservative treatment and close observation may be a viable option for SSDH presenting with TTC. However, urgent surgery may be a better option if the SSDH becomes progressively enlarged and causes compression of the spinal cord.

### ICMJE Conflict of Interest Statement

The authors declare that there is no conflict of interest that could be perceived as prejudicing the impartiality of the research reported.

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### Informed consent statement

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### CARE Checklist (2016) statement

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

### Author contribution statement

All the authors contribute to this study conduction and design. Luo performed the literature review and wrote the article.

### References


