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Cerebral venous sinus thrombosis due to external compression of internal jugular vein

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Abstract

Cerebral venous sinus thrombosis (CVST) is a special subtype of stroke that may be life-threatening in severe cases. CVST has distinct risk factors and is frequently overlooked because of its initially nonspecific clinical presentation. We herein describe a 72-year-old man who developed CVST in the right lateral sinus. Despite the absence of common risk factors in this patient, he developed external compression of the bilateral internal jugular veins by a lateral mass of the C1 vertebra and expansion of the carotid artery. Because of his elevated D-dimer and fibrinogen concentrations, which are associated with ongoing activation of the coagulation system, the patient underwent treatment with batroxobin combined with anticoagulation. Recanalization of the sinus was achieved, and his high intracranial pressure and papilledema remarkably decreased. We conclude that external compression of the internal jugular veins, which can be identified with three-dimensional computed tomography venography, may be an important risk factor for CVST.

Keywords

Lateral sinus thrombosis, venous insufficiency, batroxobin, internal jugular vein, computed tomography venography, risk factors, case report

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Introduction

Cerebral venous sinus thrombosis (CVST) is a special subtype of cerebrovascular disease in which the blocked cerebral venous outflow usually results in intracranial hypertension-related symptoms and signs. Common risk factors for CVST include pregnancy and puerperium, use of oral contraceptives, genetic thrombophilia, and infections.[1] Compared with these risk factors, internal jugular vein (IJV) abnormalities are uncommon but potentially important risk factors for CVST and have received little attention.

The current European Stroke Organization guideline recommends standard anticoagulation as the first-line treatment of CVST.[2] However, considerable numbers of patients with CVST still fail to achieve recanalization after anticoagulation treatment and thus have poor outcomes.[3] Batroxobin, a thrombin-like serine protease, may accelerate recanalization of the sinus in patients with CVST by decreasing fibrinogen and promoting thrombolysis.[4] We herein describe a 72-year-old man who developed CVST due to external compression-related bilateral IJV stenosis and underwent treatment with batroxobin combined with anticoagulation.

Case presentation

A 72-year-old man with a 2-year medical history of hypertension was referred to our clinic because of a 1-month history of progressive thunderclap headaches, visual blurriness, and nausea. His blood pressure was maintained at less than 150/90 mmHg by treatment with a beta blocker (25 mg/day of metoprolol tartrate tablets, National Medicine Standard H32025391; AstraZeneca, Cambridge, UK). The headache was described as moderate intensity, worsening in the decubitus position and occurring in the bilateral occipital regions. He reported no fever, limb weakness, or convulsions. Neurological examination was unremarkable with the exception of bilateral asymmetric papilledema, which was much worse on the right side (grade 4 in right eye and grade 2 in left eye as evaluated with the Frisén scale) (Figure 1).

The results of routine laboratory tests of the blood and cerebrospinal fluid (CSF) after admission were as follows: white blood cell count, 7.66×10^9/L; red blood cell count, 4.50×10^9/L; C-reactive protein, 8.0 mg/L; activated partial thromboplastin time, 38.3 s; international normalized ratio, 1.04; glucose level in CSF, 51.12 mg/dL; protein level in CSF, 43 mg/dL; D-dimers, 0.56 μg/mL (reference range, <0.5 μg/mL); and fibrinogen, 5.77 g/L (reference range, <4.0 g/L). The lumbar puncture opening pressure was 330 mmH_2O, and the CSF composition was normal. Ultrasonography of the carotid artery demonstrated intima-media thickening and one stable carotid plaque. Magnetic resonance venography (MRV) revealed poor visualization in the right transverse and sigmoid sinus, which was suspected to indicate thrombosis (Figure 2(a)). The thrombi in the right transverse sinus were directly displayed on the magnetic resonance black-blood thrombus imaging (MRBBTI) map (Figure 2(b)). The IJVs were visualized by three-dimensional computed tomography venography (3D-CTV) reconstruction, which revealed that the stenosis of the bilateral IJVs had been induced by a left lateral mass of the C1 vertebra and the right expanded carotid artery, respectively (Figure 2(c), (d)). In addition, optical coherence tomography revealed papilledema, abnormal retinal thickness, and macular edema with greater severity in the right eye (Figure 1(a)).

The patient received an intravenous infusion of 5 BU of batroxobin (National Medicine Standard 20031074; Beijing Topixi Pharmaceutical Co., Ltd., Beijing,

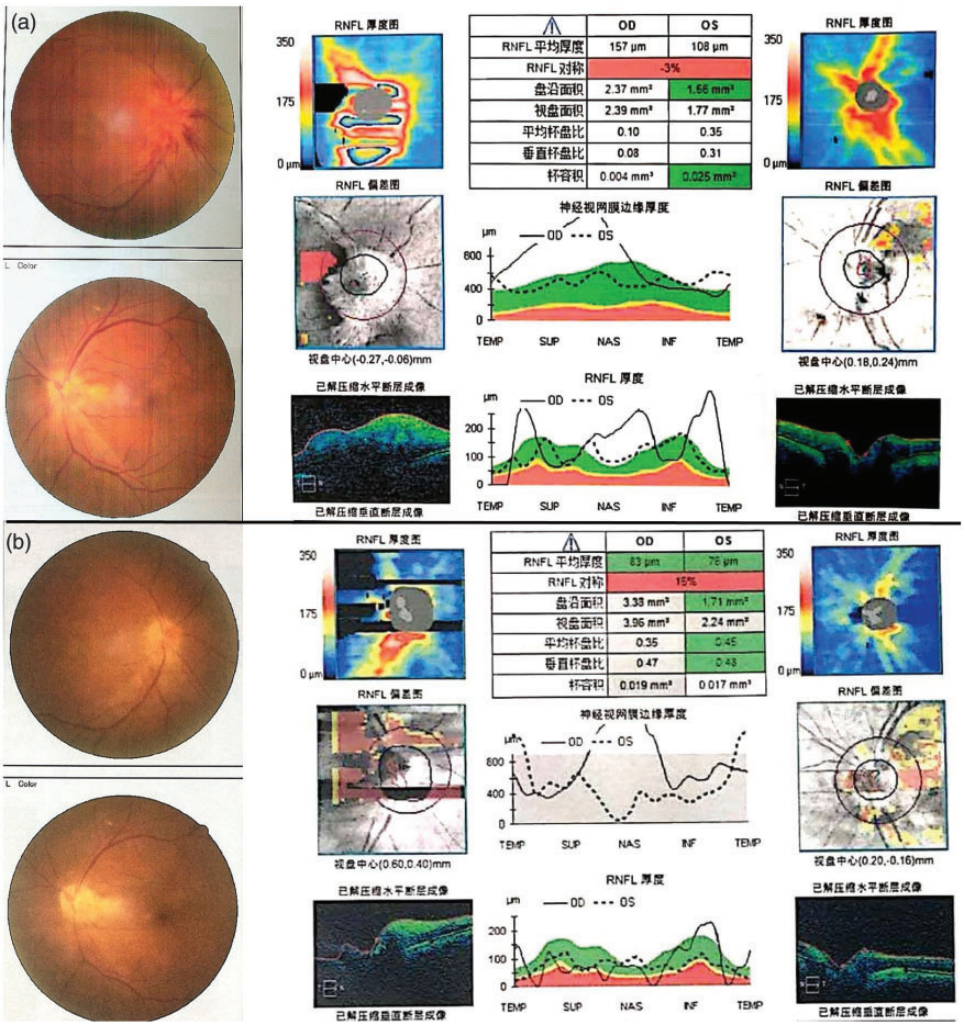

Figure 1. Fundus photography and optical coherence tomography findings. (a) Papilledema, abnormal retinal thickness, and macular edema at admission. (b) Papilledema, abnormal retinal thickness, and macular edema after 7 days of treatment.

China) every other day for a total of three doses, a subcutaneous injection of 0.6 mL of enoxaparin sodium (National Medicine Standard H20100485; Sanofi, Paris, France) every 12 hours, and an intravenous infusion of 125 mL of 20% mannitol (National Medicine Standard H34023604; Beijing Shuanghe Pharmaceutical Co., Ltd., Beijing, China) every 8 hours to alleviate his symptoms of intracranial hypertension. His symptoms gradually decreased in severity during the treatment. At the 7-day follow-up examination, his symptoms were remarkably relieved with great improvement of the papilledema (grade 1 on Frisén scale) (Figure 1(b)). The follow-up lumbar puncture opening pressure was 185 mmH_2O, and the CSF workup was normal. Moreover, both the fibrinogen level (2.07 g/L) and D-dimer

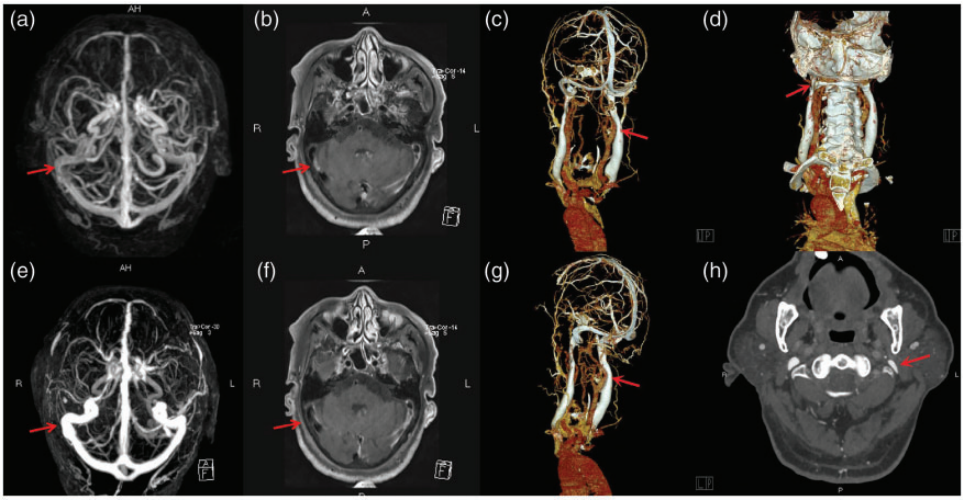

Figure 2. Cerebral venous sinus thrombosis and external compression-induced internal jugular vein (IJV) stenosis shown by magnetic resonance venography, magnetic resonance black-blood thrombus imaging (MRBBTI), and three-dimensional computed tomography venography. (a) Poor visualization in the right transverse sinus at baseline (arrow). (b) Direct thrombus signal in transverse sinus at baseline (arrow). (c) Arterial compression of J2-segment of IJVs in anteroposterior map (arrow). (d) Osseous compression of J3-segment of IJVs in anteroposterior map (arrow). (e) Follow-up magnetic resonance venography: recanalization of right transverse sinus after treatment (arrow). (f) Follow-up MRBBTI: high-intensity signal indicating diminishment of the thrombus in the transverse sinus after treatment (arrow). (g) Arterial compression of J2-segment of IJVs in oblique map (arrow). (h) Osseous compression of J3-segment of IJVs in cross-sectional map (arrow).

level (0.43 μg/mL) had decreased to within the reference range. Compared with the baseline imaging map prior to batroxobin use, the follow-up MRBBTI and MRV on post-treatment day 7 showed remarkable shrinkage of the thrombus in situ and effective recanalization of the involved sinus (Figure 2(b) versus (e) and (f)). Follow-up optical coherence tomography also indicated that the papilledema, retinal thickening, and macular edema had been remarkably attenuated, with a decrease in the Frisén grade from 4 to 1 (Figure 1).

Discussion

Our imaging examination results (3D-CTV and MRI) led to timely diagnosis of CVST. According to recent publications, physicians should be aware of warning signs of CVST (including relatively specific signs, symptoms, and risk factors) when they suspect that a patient has CVST.[5] After careful history-taking and physical examination, we identified several such warning signs, including progressive headache with aggravation in the supine position and bilateral papilledema. Magnetic resonance imaging and CTV were then performed to confirm the presence of thrombosis, and antithrombotic treatment was initiated.

Many conditions are associated with the development of CVST, and the major causes of CVST can be categorized as general and regional. Some important risk factors for CVST, such as pregnancy/puerperium, oral contraceptive use, genetic thrombophilia, and cancer, can easily result in venous thromboembolism. Other risk factors include abnormalities affecting

the head and neck region (e.g., local infection or trauma).[1] Our patient had no common risk factors, but he exhibited an abnormal regional venous structure on 3D-CTV, which appeared to be closely related to the CVST. The most common risk factors for CVST in patients of advanced age, such as our patient, are malignancy, abnormal hematological conditions, and thrombophilia, all of which are generally more relevant to aging and differ from the risk factors in young and middle-aged patients.[6,7] Notably, however, some elderly patients have no identified risk factors, suggesting that other potential risk factors or etiologies remain to be explored. In the present case, 3D-CTV revealed an overgrown left lateral mass of the C1 vertebra as well as arteriosclerosis and expansion of the right carotid artery, which led to the bilateral IJV stenosis. These are both very common conditions in the elderly population.[8,9] IJV abnormalities have also been reported to be associated with CVST, mainly thrombosis, valvular dysfunction, and stenosis.[10–12] However, the presence of multiple external compression has not been described in detail.

Extrinsic compression-induced IJV stenosis can block cerebral venous blood outflow.[13] In a report of a 15-year-old female patient, Li et al.[12] stated that CVST might be attributed to local compression of the unilateral IJV by the transverse process of the atlas and styloid process of the cranial skull; such abnormalities may originate from anatomic variations rather than the aging process. In addition, we presumed that severe aging-related degeneration of the intervertebral disks and shortening of the cervical vertebral column in the present case might have narrowed the accommodation space for cervical vessels, also potentially playing a role in blood stagnation. Thus, because specific CT sequences display not only the structure of the cerebral and jugular veins but also the relationship between blood vessels and bone, we suggest that 3D-CTV examination is needed for elderly patients with CVST, especially when contrast-enhanced MRV shows IJV stenosis, to fully explore the etiology.

Another highlight of this case is the administration of antithrombotic therapy. Batroxobin, a thrombin-like serine protease, inhibits the formation of thrombosis by inducing defibrinogenation and is extensively used to treat arterial thrombosis.[14] Our previous studies revealed that the combination of elevated D-dimer and fibrinogen concentrations may predict acute CVST and that batroxobin may safely and effectively accelerate CVST recanalization in young or middle-aged patients (age of 18–55 years), especially patients with conditions that cause hyperfibrinogenemia.[4,15] The findings in the present case, in which we achieved remarkable alleviation of symptoms and papilledema as well as sinus recanalization as shown by MRBBTI after the use of batroxobin to treat a 72-year-old patient with CVST, suggest that batroxobin combined with anticoagulation may be a promising treatment strategy in both middle-aged and elderly patients with CVST. Further clinical research is needed to explore the pathological process after batroxobin intervention.

Conclusions

Multiple external compression of the IJV, which can be identified with 3D-CTV, may be an important risk factor for CVST.

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Author contributions

GJW wrote this paper. SSY and WW provided the figures in this study, gave advice about the etiology of cerebral venous thrombosis, and

co-wrote the paper. JXM and MR contributed to the conception and design of this study and co-wrote the paper. MR takes full responsibility for the data analysis and interpretation and the conduct of the research.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics statement

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. All procedures were performed in accordance with the ethical standards of the Institutional Ethics Committee (Xuanwu Hospital, Capital Medical University). The study was approved by Ethics Committee of Xuanwu Hospital, Capital Medical University (2019-006).

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Data availability statement

The data that support the findings of this study are available from the corresponding author, MR, upon reasonable request.

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