Posterior reversible encephalopathy syndrome in a woman with pancreatitis

Jie Luo[1,2], Ya-Jing Zhan[2], Zhi-Ping Hu[2]

[1]Department of Neurology, The First People's Hospital of Changde City, Changde, Hunan 415000, China;
[2]Department of Neurology, The Second Xiangya Hospital, Central South University, Changsha, Hunan 410011, China.

To the Editor: Posterior reversible encephalopathy syndrome (PRES) is a rare condition of the central nervous system (CNS). Clinically, it can present with headache, blurred vision, seizures, or coma. The diagnosis of PRES is based on the classic radiologic imaging studies at occipital and parietal lobes in the subcortical white matter, and sometimes in the cortex. Hypertensive encephalopathy, eclampsia, immunosuppressive agents, and cytotoxic drugs can cause PRES. A patient with PRES may recover without sequela after removal of the causative factors. Notably, uncertainty in diagnosis and delay in treatment would have probably aggravated CNS injuries or death. However, the underlying mechanism of the disease remains unclear and controversial.

A 28-year-old woman was presented to the emergency department with pain in the upper abdomen, nausea, and vomiting. Her medical history included pancreatitis. Due to repeated episodes of pancreatitis, she underwent labor induction at 26 weeks of pregnancy, before she had been admitted to the hospital during her previous bout of pancreatitis. Her vital signs showed a blood pressure of 110/80 mmHg, a heart rate of 85 beats per minute, and a respiration rate of 16 breaths per minute. The physical examination was normal. Laboratory assessment was notable for amylase (3125 U/L) and lipase (100 U/L). Abdominal computed tomography (CT) scan showed an indistinct pancreatic tail with peripancreatic inflammatory changes, which demonstrated pancreatitis [Figure 1A]. One day after admission, she developed secondary infectious shock; the systolic pressure fluctuated from 50 to 59 mmHg, and diastolic pressure fluctuated from 40 to 49 mmHg. Blood pressure was normotensive after rehydration, antibiotics, and blood pressure intervention for 4 h. We then stopped using the pressor. On the fourth day, she developed diarrhea without melena. An excessive increase in blood pressure occurred (systolic pressure fluctuated from 153 to 170 mmHg and diastolic pressure fluctuated from 73 to 90 mmHg) on the fifth day. On the sixth day, she was transferred to the neurologic intensive care unit with drowsiness, seizures, blurred vision, and headache. Neurologic examination revealed cortical blindness and drowsiness. Cerebrospinal fluid cytology showed negative results for infectious etiologies. Cranial CT showed no acute intracranial hemorrhage or placeholder. Magnetic resonance venography results were normal. Magnetic resonance imaging (MRI) demonstrated areas of vasogenic edema within the cortex and the subcortical white matter of the parieto-occipital lobes [Figure 1B]. The patient made a rapid recovery after treatment with Paracillin antibiotics, somatostatin for inhibition of pancreatic fluid secretion, lansoprazole for protection of the stomach, and valproic acid sodium as an anticonvulsant. On the eighth day, she regained consciousness and was able to read the paper. Follow-up MRI at the 18th day showed complete resolution of the previously described images [Figure 1C]. These neuroimaging and clinical findings were consistent with PRES. The ethics review board of The Second Xiangya Hospital approved the study.

PRES is a rare condition of the CNS. Radiologic presentations usually involve occipital and parietal lobes, subcortical white matter, and occasionally the cortex. However, the underlying mechanism of the disease remains unclear and controversial. Two hypotheses have been put forth: damage to the autoregulation of cerebral vessels or endothelial dysfunction. The former hypothesis postulates that severe hypertension exceeds the upper limit of cerebral blood flow autoregulation and causes hyperperfusion, which may result in blood-brain barrier break down. This may then allow the interstitial extravasation of plasma and macromolecules, causing vasogenic edema. This hypothesis could explain the pathophysiology of PRES in patients with severe hypertension, which exceeds the upper limit of autoregulation at mean arterial pressures (MAPs) greater than 150 to 160 mmHg. However, this

Access this article online

Quick Response Code:

Website: www.cmj.org

DOI: 10.1097/CM9.0000000000000431

Correspondence to: Dr. Ya-Jing Zhan, Department of Neurology, The Second Xiangya Hospital, Central South University, Renmin Road 139, Changsha, Hunan 410011, China
E-Mail: zhanyajing@csu.edu.cn

Chinese Medical Journal 2019;132(18)
Received: 25-03-2019 **Edited by:** Yi Cui

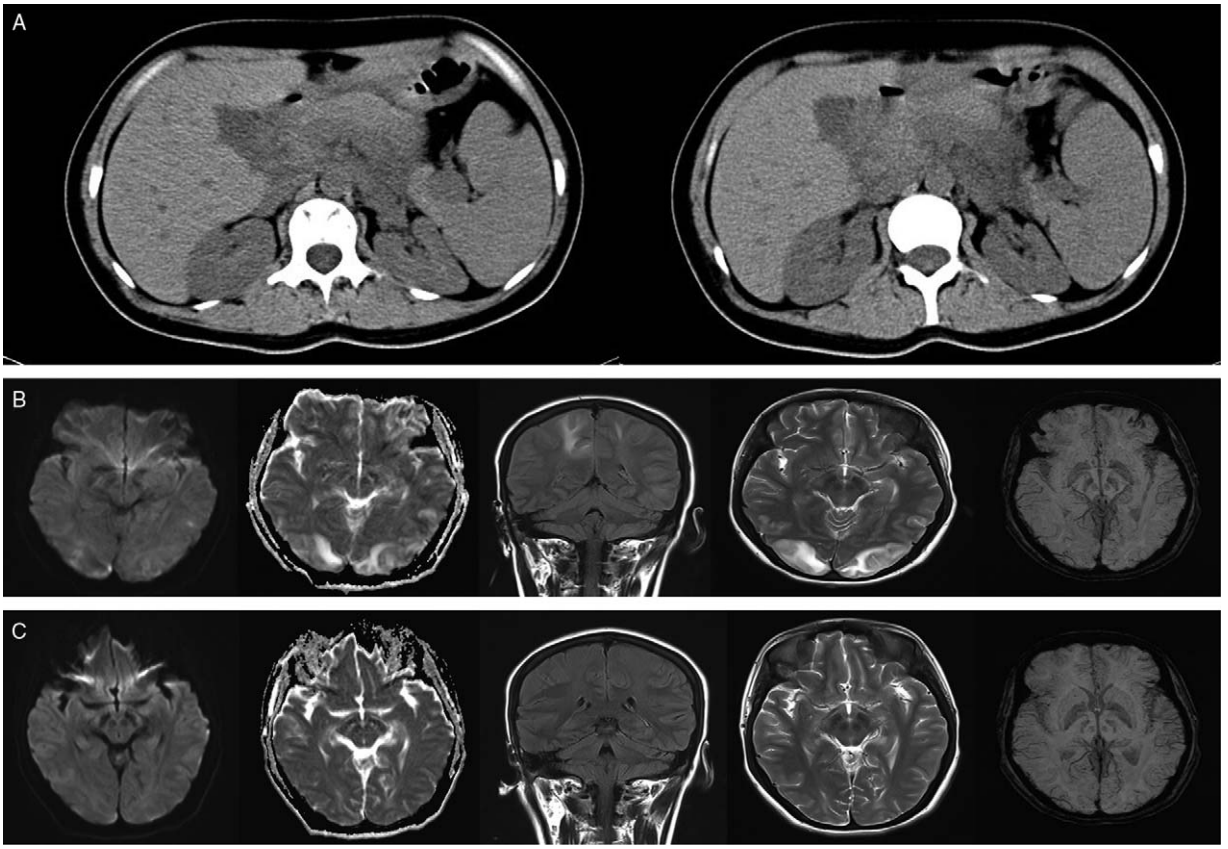

Figure 1: Computed tomography (CT) and magnetic resonance imaging of the patient. (A) Abdominal CT showed the pancreatic tail was swollen and indistinct. (B) The diffusion-weighted image of the posterior cerebral white matter was the equal and the partial high signal, the high signal of the apparent diffusion coefficient (ADC), which conforms to vasogenic edema. Susceptibility-weighted imaging demonstrated no obvious microhemorrhage. (C) Magnetic resonance imaging performed 18 days later, and nidus was further reduced, which only remained the right posterior occipital lobe flecked lesions after symptomatic support treatment.

theory does not explain the pathophysiology for patients with moderate hypertension below this MAP limit. The latter hypothesis proposes that cytotoxic drugs, cytotoxins, metabolites, chemokines, and cytokines (eg, tumor necrosis factor-α, interleukin-1, and interferon-γ) may cause endothelial dysfunction that can lead to vasoconstriction of microvasculature, which may be further aggravated by hypertension and associated automatic regulation of cerebral vascular response. Finally, such vasoconstriction may cause resultant hypoperfusion and ischemia. Possible etiologies for PRES are varied and include preeclampsia, eclampsia, hypertensive encephalopathy, underlying autoimmune conditions, and the use of cytotoxins or immunosuppressants.

We reported a unique case of a patient with typical clinical and radiologic findings of PRES after having pancreatitis. Reviewing similar cases in the medical literature, few cases of PRES have been shown to follow pancreatitis.[1,2] From these cases, we could conclude that acute pancreatitis combined with PRES mostly occurs in patients with moderate hypertension. When headache, visual impairment, and epilepsy occur in a patient with acute pancreatitis, PRES should be considered. In acute pancreatitis, a local and systemic activation of inflammatory pathways could injure secondary intracranial vascular endothelium, causing cytotoxic and vasogenic edema. Experimental models of pancreatitis have shown that the proinflammatory cytokines produced during pancreatitis may have a pivotal role in cytotoxic and vasogenic edema, which may be a pathogenic mechanism similar to PRES.[3] In our previous review of the literature, we found no case report of acute pancreatitis in a retrospective study of 411 patients with acute ischemic stroke, whereas another study reported one cerebral hemorrhage patient with complications of pancreatitis.[4,5] The main differences from previously reported cases include following. First, except for pancreatitis, most of the previously reported patients had a history of leukemia or heavy drinking, but our case only had a history of pancreatitis. Second, our patient had a history of recurrent pancreatitis. In this case, the automatic regulation of cerebral vessels hypothesis does not account for the full spectrum of pathophysiology of this disorder, as with other patients with moderate hypertension. After recurrent pancreatitis, hyperactivation of trypsinogen and the release of various inflammatory mediators, cytokines, and chemokines likely cause subsequent endothelial dysfunction that can lead to resultant hypoperfusion and ischemia. Thus, we suggest that pancreatitis may be an etiology of PRES. Acute pancreatitis is a common clinical disorder presenting as an acute abdomen. While pancreatitis itself can be life-threatening, this case reminds clinicians of unusual complications. In pancreatitis patients with classic neurologic symptoms, the possibility of PRES should be considered regardless of the timing of the onset of symptoms.

Declaration of patient consent

The author certifies that they have obtained all appreciate patient consent forms. In the form, the patient/patient's guardians have given their consent for their images and other clinical information to be reported in the journal. The patient/patient's guardians understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Funding

This study was supported by the grants from the National Natural Science Foundation of China (No. 81801135) and the Hunan provincial Science Foundation (No. 2017JJ3477).

Conflicts of interest

None.

References

1. Baek HS, Lee SJ. A case of posterior reversible encephalopathy syndrome associated with acute pancreatitis and chronic alcoholism. Gen Hosp Psychiatry 2015;37:192.e3-5. doi: 10.1016/j.genhosppsych.2014.12.004.
2. Compagnone C, Bellantonio D, Pavan F, Tagliaferri F, Barbagallo M, Fanelli G. Posterior reversible encephalopathy syndrome in acute pancreatitis. Minerva Anestesiol 2016;82:1236–1237. Epub 2016 Jun 7.
3. Farkas G, Marton J, Nagy Z, Mandi Y, Takacs T, Deli MA, *et al*. Experimental acute pancreatitis results in increased blood-brain barrier permeability in the rat: a potential role for tumor necrosis factor and interleukin 6. Neurosci Lett 1998;242:147–150. doi: 10.1016/s0304-3940(98)00060-3.
4. Ota K, Oniki A, Kobayashi Z, Ishihara S, Tomimitsu H, Shintani S. Acute pancreatitis is a very rare comorbidity of acute ischemic stroke. J Rural Med 2018;13:72–75. doi: 10.2185/jrm.2956.
5. Zhong X, Gong S. Fatal cerebral hemorrhage associated with acute pancreatitis: a case report. Medicine (Baltimore) 2017;96:e8984. doi: 10.1097/MD.0000000000008984.

How to cite this article: Luo J, Zhan YJ, Hu ZP. Posterior reversible encephalopathy syndrome in a woman with pancreatitis. Chin Med J 2019;132:2265–2267. doi: 10.1097/CM9.0000000000000431