Homonymous quadrantopia associated with hyperosmolar hyperglycemic syndrome

Hemianopia occurs as a rare complication of diabetic hyperglycemic hyperosmolar syndrome (HHS; Table S1)1-4, but quadrantopia has not been reported as a complication of diabetes.

A man aged in his 60s presented to us with a 3-day history of reddish and greenish hallucinations with impaired focusing. He had also experienced a mild frontal headache for a few days. He was diagnosed as having diabetes approximately 10 years earlier with glycosylated hemoglobin >10% (86 mmol/mol). He reported receiving a non-pharmacological treatment, and his glycosylated hemoglobin level lowered to approximately 6.0% (42 mmol/mol) according to him. However, he had left the diabetes untreated in the recent 5 years. On presentation, he was fully alert and complained of excessive thirst. He was obese (the body mass index 27.9 kg/m²), and a physical examination revealed dry mouth; his blood pressure was 138/82 mmHg and physical findings were otherwise not contributory. His random sample plasma glucose was 32.0 mmol/L, glycosylated hemoglobin 14.8% (138 mmol/mol), osmolality 302 mOsm/kg, urinary sugar was 3+, ketone bodies negative and plasma 3-hydroxybutyrate 75 µmol/L (reference range <85 µmol/L).

The patient was fully alert and had no involuntary movements, such as choreoathetosis, ballismus, dysphagia and seizures, so we did not carry out electroencephalography. Ophthalmological examination showed discrete homonymous right inferior quadrantopia (Figure 1, upper panel) and mild non-proliferative diabetic retinopathy. The anopia mostly dissipated by day 8 (Figure 1, lower panel), with lowering of plasma glucose during this period to 10.7 ± 2.5 mmol/L (mean ± standard deviation of 32 blood samples, of which 21 were preprandial samples and 11 post-prandial samples) by fluid and insulin. Magnetic resonance imaging (MRI) of the brain taken on day 1 and 8 showed no abnormality (Figure S1). The patient returned to normal daily life on day 10, and there have been no visual symptoms after the discharge, during 6 months of follow up.

This is the first documentation of quadrantopia associated with HHS. The quadrantopia had a rapid onset and was short-lasting, and well synchronized with the temporal profile of glycemia in this patient. Sharply demarcated inferior quadrantopia seen in the present patient suggested the localization of the responsible neuronal lesion in the occipital lobe. However, the brain MRI was normal throughout the course of the current episode. Vascular events and brain tumors might be listed as differential diagnosis, but normal MRI easily excluded such possibilities. There have been case reports in which HHS-associated hemianopia without MRI abnormality was documented1-4. The mechanisms of cerebral injury in quadrantopia associated with hyperglycemia are unclear. For hemianopia, it was postulated to include intracellular dehydration, reactive oxygen species generation and others by Strowd et al.4 Some of them might have been responsible for the quadrantopia seen in the present patient. Whatever the abnormality was, the rapid recovery and negative MRI suggested that the lesion might have been small in size and/or the degree of plasticity rather high. Diagnosis of HHS at a relatively mild stage followed by rapid normalization of glucose might have contributed to the benign clinical course of this patient as well.

In conclusion, one should keep in mind that hemianopia and quadrantopia can take place as neuro-ophthalmological complications of HHS.

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DISCLOSURE
The authors declare no conflict of interest.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Figure S1 | Brain magnetic resonance imaging (T2-weighted image). There are no abnormalities, such as occipital subcortical hypointensity, in these images. No abnormality was observed in the optic tract as well (not shown).

Table S1 | Previous cases of hemianopia associated with hyperglycemia.