Recurrent painful ophthalmoplegia in a patient with diabetes mellitus: Is ophthalmoplegia associated with diabetes mellitus?

Dear Editor:

Tolosa-Hunt syndrome (THS) and diabetes mellitus (DM) are relatively common etiologies of painful ophthalmoplegia (PO). However, distinguishing diabetic ophthalmoplegia from THS or recurrent painful ophthalmoplegic neuropathy (RPON) is sometimes difficult because they demonstrate similar clinical courses and DM existence does not necessarily confirm the diagnosis of diabetic ophthalmoplegia (1).

We encountered a case of a 49-year-old man with DM and recurrent PO affecting alternative sides, exhibiting clinical features similar to THS; he complained of severe pain in the right periorbital and temporal regions that occurred two days prior to diplopia and right ptosis. He had been diagnosed with DM seven years before and was treated with 3 mg glimepiride and 500 mg metformin per day without any diabetic complications. He had neither past medical nor family history of headache. Neurological examination showed an outward deviation of the right eye with partially impaired movement and right ptosis. Pupils were isochoric; light reflexes were prompt in the eyes. Laboratory findings showed elevated levels of hemoglobin A1C of 10.0% and 239 mg/dl blood glucose. Computed tomography (CT) and contrast-enhanced magnetic resonance imaging (MRI) of the head showed intact brain parenchyma and craniofacial structures, including sinuses, mastoids and orbits. After excluding vascular disease, neoplasm, inflammation and infection, he was treated with oral prednisolone, resulting in complete pain disappearance on the following day. Right ophthalmoplegia and ptosis improved and completely disappeared after two months.

However, after one month, the patient was readmitted because of severe pain in the left periorbital and temporal regions that occurred a day prior to diplopia and left ptosis. Neurological findings were mostly similar to those at prior admission, except that the affected side changed from right to left. Laboratory findings were unremarkable, except for elevated levels of hemoglobin A1C of 8.9% and 318 mg/dl blood glucose. CT and contrast-enhanced MRI of the head were inconclusive. Considering that recurrence ensued over a relatively short period, he received intravenous and oral prednisolone. His pain completely disappeared the following day; left ophthalmoplegia and ptosis improved and completely disappeared after two months, as in the past attack.

Although the patient had poorly controlled DM, his clinical course characterized by ophthalmoplegia with ipsilateral severe headache and rapid response to steroid therapy was similar to that of THS. THS was clinically diagnosed, but the possibility of diabetic ophthalmoplegia could not be completely excluded.

Because THS etiology remains unknown, the International Headache Society’s diagnostic criteria differ in each edition (1). However, the latest diagnostic criteria require abnormal findings on MRI or biopsy (2). Zhang et al. doubted the validity of these criteria and indicated the need for proper change (1) because some cases are clinically indistinguishable from THS with normal MRI and are more recently named as benign THS (3). The borders between diabetic ophthalmoplegia, THS with DM, and RPON with DM remain unclear because their etiologies and diagnostic criteria are uncertain. Because only a few studies clarified various clinical features and pathological mechanisms of PO with DM, clinical studies by headache experts focusing on PO with DM are needed to better define classifications and diagnostic criteria.

Declaration of conflicting interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The authors received no financial support for the research, authorship, and/or publication of this article.

Acknowledgment

The authors would like to thank Dr. Masanori Kurihara for generous support and Enago (www.enago.jp) for the English language review.
References


Kensuke Hamada, Yasuhsa Sakurai and Izumi Sugimoto
Department of Neurology,
Mitsui Memorial Hospital, Japan

Corresponding author:
Kensuke Hamada, Department of Neurology,
Mitsui Memorial Hospital, 1, Kanda-Izumi-cho,
Chiyoda-ku, Tokyo 101-8643, Japan.
Email: khamada-tky@umin.ac.jp