Lymphocytic Adenohypophysitis Accompanying Occlusion of Bilateral Internal Carotid Arteries

—Case Report—

Jun-ichi IKEDA, Jun-ichi KURATSU, Masaki MIURA, Yutaka KAI and Yukitaka USHIO

Department of Neurosurgery, Kumamoto University Medical School, Kumamoto

Abstract

A 45-year-old female presented with blurred vision, headache, and abnormal thirstiness. She was not pregnant nor postpartum. She had diabetes insipidus and bitemporal hemianopsia. Radiological evidence suggested a mass arising in the sella turcica with extension into the parasellar and suprasellar regions. Carotid angiography showed occlusions of bilateral internal carotid arteries at the cavernous portions. The mass was subtotaly removed by the trans-sphenoidal approach and was histologically diagnosed as an adenohypophysitis. Laboratory data showed the patient to be in an active state of autoimmune disorder and hypopituitarism. When a patient presents with pituitary insufficiency and an enhanced intrasellar mass lesion on computed tomographic scan, lymphocytic adenohypophysitis must be included in the differential diagnosis.

Key words: adenohypophysitis, autoimmune, pituitary, internal carotid artery occlusion

Introduction

Lymphocytic adenohypophysitis is a rare disorder reported almost exclusively in pregnant or postpartum females.\(^6\) At present, the pathogenesis is obscure but it has been considered an autoimmune disorder.\(^6\) We report a 45-year-old female, neither pregnant nor postpartum, with lymphocytic adenohypophysitis. Laboratory data suggested she was in an active state of autoimmune disorder and carotid angiography showed occlusion of bilateral internal carotid arteries (ICAs) at the cavernous portion. To the best of our knowledge, this is the first reported case of ICA occlusion by lymphocytic adenohypophysitis.

Case Report

A 45-year-old female was admitted to our service on September 28, 1987 with a 3-year history of blurred vision, headache, and abnormal thirstiness. On admission, she was obese, weighing 75 kg and was 152 cm in height. Neurological examination revealed a bitemporal upper quadrant hemianopsia. Preoperative hormonal examinations showed serum prolactin (PRL) 16.0 ng/ml (normal range, 2.0–30.0), growth hormone (GH) 1.0 ng/ml (less than 5.5), luteinizing hormone (LH) 3.6 mIU/ml (19–134), follicle-stimulating hormone (FSH) 2.1 mIU/ml (26–149), thyroid stimulating hormone (TSH) 1.3 µU/ml (0.36–3.25). Serum cortisol was under 2.0 µg/ml. The thyroxine (T4) radioimmunoassay revealed 2.4 µg (normal, 4.5–12), triiodothyroxine (T3) uptake was 1.1 µg/ml (0.7–2.1). The immunological examinations showed OKT4 44% (35.5–46.9), OKT8 12.5% (21–32), OKT4/OKT8 3.52 (0.6–2.9), CH50 39 U/ml (30–40), C3 71 mg/ml (70–110), C4 16 mg/ml (22–40). She was positive for antinuclear antibodies.

Plain skull x-ray showed a saucer like deformity of the sella turcica and intrasellar calcification (Fig. 1). Computed tomographic (CT) scans revealed an isodense mass in the sella and suprasellar region; it was homogeneously enhanced by the contrast medium (Fig. 2). Magnetic resonance (MR) imaging demonstrated the lesion as an isointense mass on T1-weighted image (Fig. 3) and as a slightly high-intense mass on T2-weighted image. Left carotid
angiography showed complete occlusion of the bilateral ICAs at the level of the cavernous portion (Fig. 4). The bilateral cerebral hemispheres were fed by verteobasilar arteries. The bilateral external carotid arteries were well-developed and also fed the cerebral hemispheres.

A diagnosis of pituitary adenoma or craniopharyngioma was made and she underwent trans-sphenoidal exploration of the sella on October 20, 1987. Firm, yellow tissue in the sella was partially removed. Histological examination of the surgical specimens showed an abnormal anterior pituitary gland with extensive fibrosis and scattered small nests of pituitary cells. There was infiltration of the tissue by numerous lymphocytes and plasma cells (Fig. 5). Immunohistological sections showed cells containing PRL, GH, adrenocorticotropic hormone, TSH, FSH, and LH.

**Discussion**

Lymphocytic adenohypophysitis is a distinct and specific pathological disease process whose pathogenesis is unclear at present. With the exception of one patient, all previously reported patients were pregnant or postpartum. Therefore, lymphocytic adenohypophysitis has been considered to be closely related to pregnancy. We report the first case of lymphocytic adenohypophysitis in a female who was neither pregnant nor postpartum. Furthermore, our patient presented with occlusion of the bilateral
ICAs. Autoimmune disorder is believed to play an important role in the development of lymphocytic adenohypophysitis. Bottazzo et al.\(^3\) identified autoantibodies to PRL-secreting cells in about 7% of patients having one or more autoimmune endocrine diseases. Engelberth and Jezkova\(^3\) found these autoantibodies in 18% of females during the first week after childbirth. Shanklin\(^3\) demonstrated at autopsy that 43% of apparently normal pituitaries were infiltrated by lymphocytes. Pituitary lymphocytic infiltration and antipituitary antibodies are a frequent and specific finding in autoimmune disorder but the relationship to lymphocytic adenohypophysitis remains unclear. Our patient lacked clinical evidence of associated autoimmune endocrinopathies, but laboratory data showed complementary consumption (decrease of serum C4) which is suggestive of an active immunological state and the presence of antinuclear antibodies which is indicative of autoimmune disease. Vanneste and Kamphorst\(^3\) suggested that a history of lymphocytic meningitis may be a contributing factor in lymphocytic adenohypophysitis.

In our case, angiography showed the occlusion of bilateral ICAs at the level of the cavernous portion. ICA occlusion by an intracranial tumor is rare. Meningioma and adenoma were the most common tumors in patients with ICA occlusion.\(^{12,16,19}\) Radiation therapy and neurocutaneous syndrome may be contributory factors in patients with carotid occlusion.\(^6\) However, our patient had not received radiation therapy nor did she present with neurocutaneous syndrome. Therefore, we considered the possibility of lateral extension of lymphocytic adenohypophysitis. Her clinical finding suggested that the obstruction of the artery was slowly progressive and well compensated. An angiogram showed a good collateral flow to the affected hemisphere, which accounted for the lack of ischemic symptoms. To the best of our knowledge, this is the first reported case of ICA occlusion by lymphocytic adenohypophysitis.

Recently, Levine et al.\(^{13}\) reported that the signal intensity of lymphocytic adenohypophysitis by T1-weighted MR image was isointense, quite similar to that of pituitary adenoma. In our case, the MR imaging demonstrated isointensity on the T1-weighted image and slight high intensity on the T2-weighted image. Therefore, it seems to be difficult to differentiate distinctly between pituitary adenoma and lymphocytic adenohypophysitis by MR imaging.

When a patient presents with pituitary insufficiency and a sellar and/or suprasellar enhanced lesion is visible on CT scan, lymphocytic adenohypophysitis must be included in the differential diagnosis, especially when the findings on the CT scan contrast with the normal radiography of the sella.

References


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Address reprint requests to: J. Ikeda, M.D., Department of Neurosurgery, Kumamoto University Medical School, 1–1–1 Honjo, Kumamoto 860, Japan.

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