HORMONES 2017, 16(3):331-332

Letter to the Editor

A case of idiopathic granulomatous hypophysitis

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Dear Sir,

Hypophysitis is a rare disease of the pituitary gland whose incidence is 1 case per 9 million people per year.[1] Hypophysitis, which can be categorized as primary (idiopathic) hypophysitis and secondary hypophysitis, may develop through systemic inflammatory disorders such as tuberculosis, Wegener's granulomatosis and sarcoidosis.[2] Based on histologic features there are two main types of hypophysitis: lymphocytic and granulomatous. Three rare forms have been described, namely IgG4-related hypophysitis, necrotizing hypophysitis and mixed forms.[3] Granulomatous hypophysitis, first described in 1917 by Simmonds,[4] has an incidence of 1 in 10 million and constitutes less than 1% of all pituitary disorders.[5] Unlike lymphocytic hypophysitis, there is no sex preference. The mean age of diagnosis is 20 yr for females and 50 yr for males. This disorder is characterized by necrotizing granulomas that are formed by histiocytes and plasma cells surrounding areas of necrosis.[6] It is easily initially diagnosed as a pituitary adenoma, this based on nonspecific radiological features. We would like to report a rare case of idiopathic granulomatous hypophysitis presenting as a sellar mass and associated with hypopituitarism.

Key words: Idiopathic granulomatous hypophysitis, Magnetic resonance imaging, Pituitary adenomas

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Received:10-10-2017, Accepted:12-10-2017

A 34-year-old male visited the endocrine clinic complaining of headache, fatigue and loss of libido. On physical examination his blood pressure was 120/70 mmHg and his pulse rate was 64 beat/min. Central obesity was present. The results of his hormone tests were as follows: TSH: 1.27μIU/ml, fT4: 0.582ng/dl, fT3: 1.88pg/ml, ACTH: 14.8pg/ml, cortisol: 9.05ug/dl, LH: 0,81mIU/ml, FSH: 0.97mIU/ml, prolactin: 22.23ng/ml, GH: 0.086ng/ml, IGF-1: 98 (115-307) ng/ml. His urine density was 1013. After the detection of hypopituitarism, a hypophysis magnetic resonance imaging (MRI) was requested. The MRI showed a macroadenoma located in the adenohypophysis. The tumor, measuring 15×9×10mm, had pushed the normal parenchyma inferiorly pressing it into a tape-like form and the infundibulum slightly towards the superiorly (Figure 1). The patient first received steroid treatment followed by thyroid hormone replacement treatment and was referred to the neurosurgery department. A total adenoma excision was performed using the endoscopic endonasal transsphenoidal technique. The pathology test result revealed lymphoplasmacytic cell infiltration that had extensively damaged the adenohypophysis and non-necrotizing granuloma containing Langhans-type multinucleated giant cells. Gomori metanamin silver, periodic acid-Schiff and asidoresistant bacilli

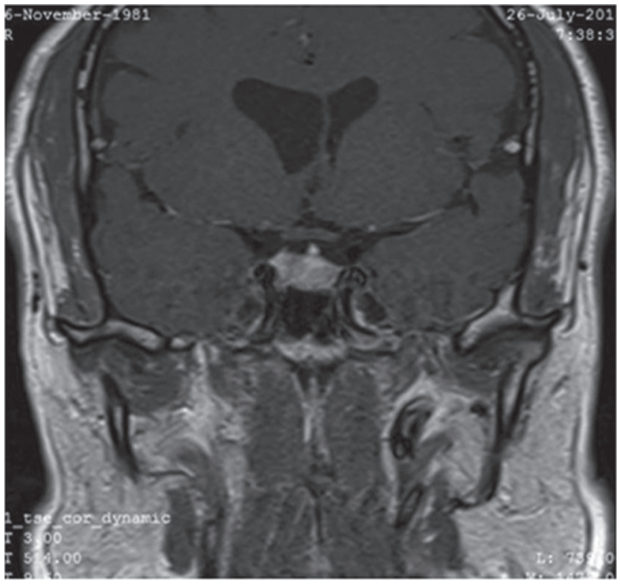

Figure 1. The MRI showed a macroadenoma located in adenohypophysis. The tumor, 15×9×10mm, has pushed the normal parenhyma inferiorly, pressing it into a tape-like and the infundibulum slightly towards syperiorly.

specific organisms were not observed. The pathological diagnosis was determined as granulomatous hypophysitis. The patient is on hydrocortisone 20mg/day and levothyroxine 75mcg/day and is receiving testosterone replacement every 3 weeks.

The clinical appearance of hypophysitis may mimic that of a pituitary adenoma.[7] It includes headache, nausea, fatigue, hypopituitarism and diabetes insipidus. If there is mass effect on the optic chiasm vision may be distorted, usually with a typical bitemporal hemianopsia, while if the cavernous sinus is affected patients may have diplopia. Granulomatous hypophysitis is a very rare pituitary condition that is difficult to diagnose before surgery. The diagnosis of idiopathic granulomatous hypophysitis (IGH) is made following exclusion of secondary causes.[8,9]

MRI-specific information on IGH is sparsely reported in the literature, with pituitary enlargement being the most common feature. Contrast enhancement and pituitary stalk thickening can also be seen. Clinically and radiologically, IGH is a rare sellar structure that can be easily misdiagnosed as a pituitary adenoma.[10]

There is strong evidence in the published literature that anti-pituitary autoantibodies most probably play a role in the pathogenesis of autoimmune pituitary disorders.[11]

In conclusion, hypophysitis is a heterogeneous group of inflammatory conditions involving the pituitary gland. Idiopathic granulomatous hypophysitis is a very rare hypophyseal condition that is difficult to diagnose before surgery. The diagnosis of idiopathic granulomatous hypophysitis is made after excluding secondary causes. Despite medical treatment of the majority of suspicious cases, an increase in the size of the gland and symptoms associated with mass effect may lead to surgical intervention and gland resection.

CONFLICT OF INTEREST

The author does not have any conflict of interest.

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