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Case Report

Growth hormone-secreting adenoma coexisted with gangliocytoma: a rare case

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Abstract: A gangliocytoma in the sellar region is extremely rare. We report a rare case of mixed gangliocytoma and growth hormone (GH)-secreting adenoma in a 50-year-old woman, who presented with acromegaly. Laboratory investigations revealed high levels of GH and insulinlike growth factor 1 (IGF-1). Sellar computed tomography scan and contrast enhanced magnetic resonance imaging (CE-MRI) showed a sellar mass. Based on clinical, biochemical, and radiologic evaluations, GH-secreting adenoma was diagnosed and operated by endonasal transsphenoidal approach achieving total removal of the tumor. After surgery, histopathological examination demonstrated mixed gangliocytoma and GH-secreting adenoma in the resected lesion. The clinical, radiological, and operative data are reviewed, as are the histological findings. To our knowledge, few cases of mixed gangliocytoma and GH-secreting adenoma have been reported.

Keywords: Pituitary adenoma, gangliocytoma, growth hormone

Introduction

Pituitary adenoma is the most common type of tumor found in the sellar region, which account for 10-15% of all primary brain tumors. Gangliocytomas in the seller are uncommon entities, the natural history of which is not understood. There have been few cases of intrasellar gangliocytoma since the first report by Greenfiled in 1919 [1]. And growth hormone (GH)-secreting adenoma combined with gangliocytoma is extremely rare. The most common syndromes are acromegaly and precocious puberty. We report a rare case of GH-secreting adenoma combined with gangliocytoma in a 50-year-old woman.

Case report

A 50-year-old woman was admitted to our hospital presented with intermittent headache. A general physical examination disclosed acromegaly (**Figure 1A**). And her blood pressure and glycated hemoglobin was normal. Laboratory evaluation revealed high-level GH about 19.83 ng/ml and insulinlike growth factor 1 (IGF-1) of 967 ug/L. Magnetic resonance imaging (MRI) revealed a slightly T1-hyperintence and T2-hypointence mass with T2-hyperintence in its center (**Figure 1B** and **1C**); contrast enhanced MRI (CE-MRI) showed a sellar obviously enhancing mass about 2.1×1.9×1.8 cm after gadolinium administration which had a circular boundary and push the optic chiasma to the upward side (**Figure 1D-F**). A clinical diagnosis of GH-secreting adenoma was made and she was operated by the transsphenoidal approach under microscope. A total resection of the tumor was achieved. And the patient's serum GH level returned to normal 2 days after the operation. And 6 months later, CE-MRI confirmed the total resection of the tumor (**Figure 3**).

The histopathological examination revealed the presence of two main components in the tumor: pituitary adenoma cells and ganglion cells (**Figure 2A**). And the ganglion cells lies in plenti-

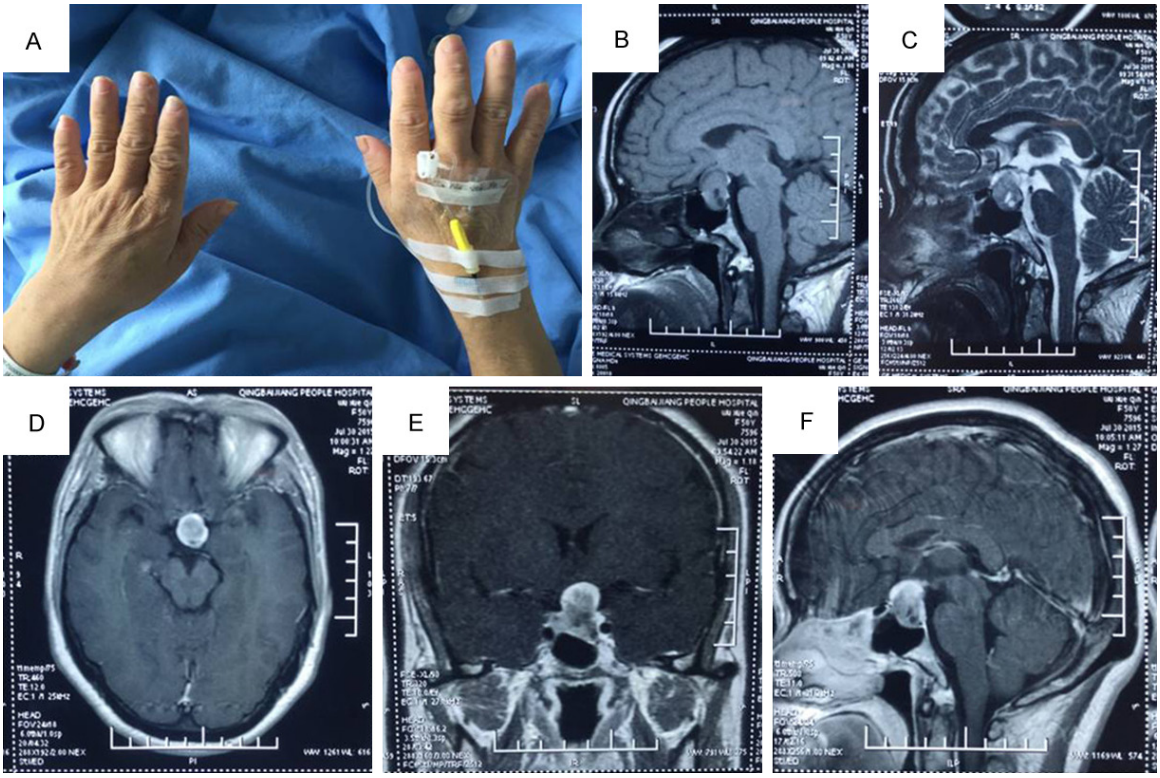

Figure 1. Physical examination found thick fingers with enlarged interphalangeal joints on the both side (A); MRI revealed a slightly T1-hyperintence and T2-hypointence mass with T2-hyperintence in its center (B and C); CE-MRI disclosed a sellar obviously enhancing mass about 2.1×1.9×1.8 cm after gadolinium administration which had a circular boundary and push the optic chiasma to the upward side (D-F).

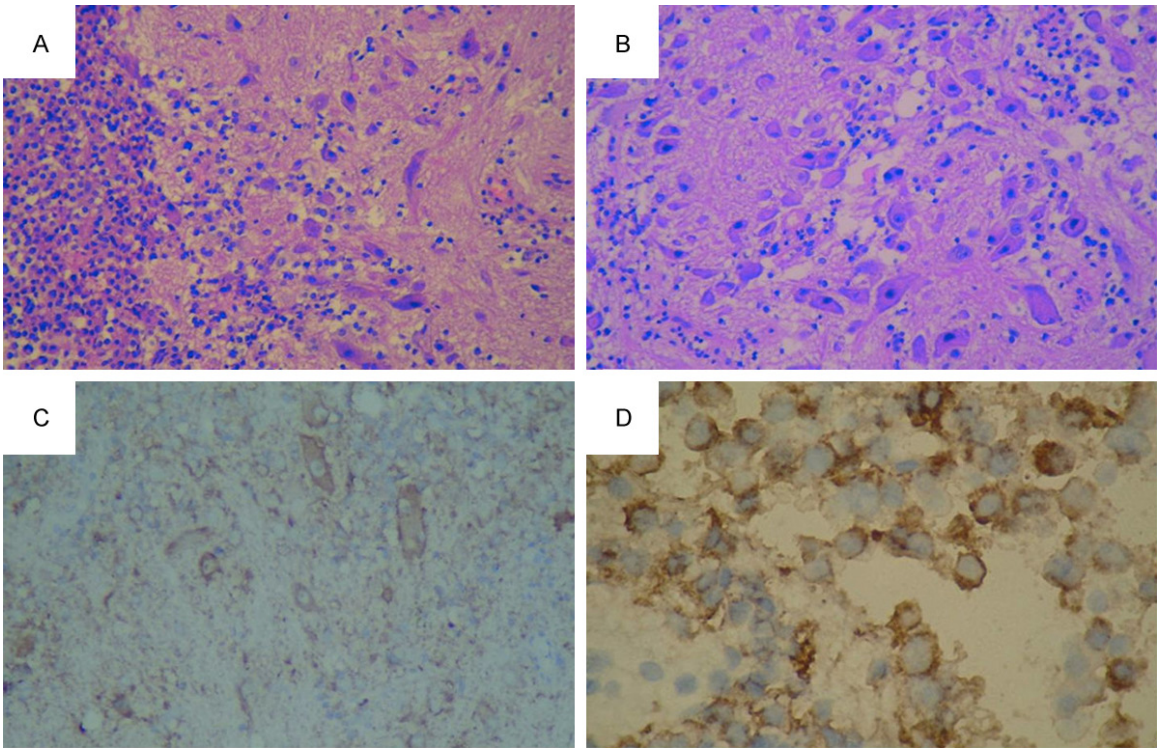

Figure 2. Ganglion cells and pituitary adenomatous cells. No sharp border exists between the two components (HE staining, original magnification, ×100) (A). The ganglion cell area. The ganglion cells lie in abundant neuropil, with

perivascular lymphocytic infiltration (HE staining; original magnification, ×200) (B). Ganglion cells immunoreactive for neuron-specific enolase (diaminobenzidine and Meyer's haematoxylin, original magnification, ×200) (C). Pituitary adenomatous cells immunoreactive for growth hormone (diaminobenzidine and Meyer's haematoxylin, original magnification, ×400) (D).

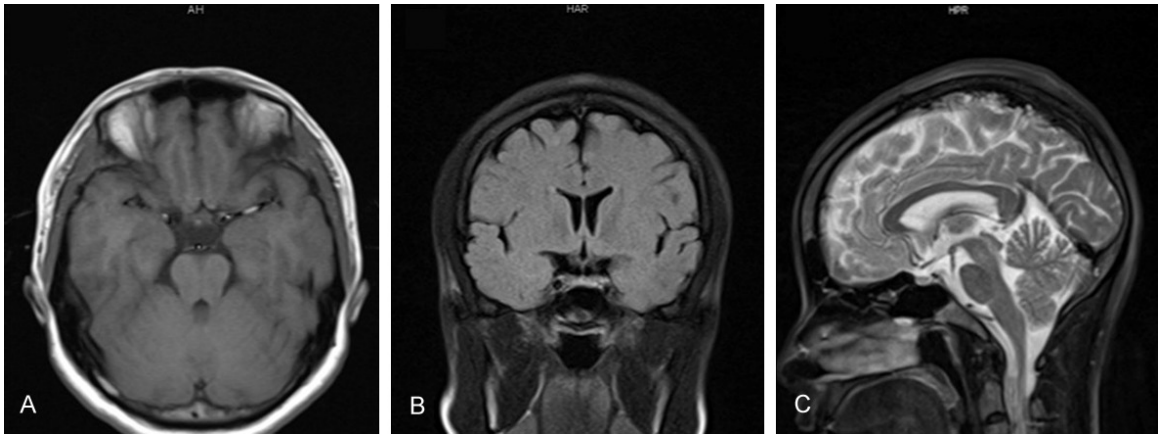

Figure 3. 6 months after the operation. MRI shows total removal of the tumor (A-C).

ful neuropil with lymphocytic infiltration (**Figure 2B**). Adenoma cells were immunareactive for GH and ganglion cells were positive for neuron-specific enolase (**Figure 2C** and **2D**).

Discussion

Pituitary adenoma is a common benign tumor in the sellar region while gangliocytoma is a rare benigh tumor originating from the posterior pituitary. And growth hormone (GH)-secreting adenoma combined with gangliocytoma is extremely rare. A believable hypothesis is that the gangliocytoma originates from the neural differentiation of a pre-existing pituitary adenoma [2, 3].

The most common symptom is acromegaly and only a few tumors have been reported to be non-functioning [4]. And surgical resection is often recommended for this mixed gangliocytoma and pituitary adenoma that present with visual defects or endocrinopathy. And radiotherapy is not indicated in the management of the tumors because the therapy efficacy of it on the slowly growing tumors is uncertain.

Several interesting theories have been used of the mixed tumor including the following: (1) Metaplasia theory, one tumor is the original tumor while the others is the metaplastic tumor; (2) Induced theory, one tumor occurs first and induces the occurrence of the other type of tumor; (3) Encounter theory, two tumors of different origins occur respectively and mix together; (4) Gene theory, tumorigenic factors act on the oncogenes of adenohypophysis and neurohypophysis, and the activate cell genesis synchronously; (5) Stem cell differentiation theory [5, 6].

In summary, these mixed gangliocytoma and pituitary adenoma have rarely been reported worldwide. Since the mixed tumors are not capable to metastasize, the therapeutic effects may be satisfied by surgery alone. Therefore, this report may help neurological surgeons to a better understanding of mixed pituitary tumors.

Disclosure of conflict of interest

None.

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