

One exceedingly rare co-existence of pituitary adenoma with hydrocephalus and cerebral aneurysm: case report and literature review

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ABSTRACT: The author described an exceedingly uncommon case of a giant pituitary adenoma with hydrocephalus and cerebral aneurysm simultaneously. A 59-year-old man had a 1-month history of headache, dizziness, and visual disturbance. Preoperative evaluation showed a pituitary tumor and hydrocephalus. A cerebral aneurysm was found at the anterior communicating artery during the surgery (transcranial and transventricular approach). The aneurysm was clipped and the pituitary tumor was completely removed. Postoperative resolution of hydrocephalus was confirmed on MRI scan at 3 months after surgery, and satisfactory clipping of the aneurysm was confirmed on 64-slice CT angiogram (CTA). Pituitary adenomas causing hydrocephalus are rare, and literature review showed no previous report of a giant pituitary adenoma with hydrocephalus associated simultaneously with a cerebral aneurysm. To our knowledge, this is the first report of giant pituitary adenomas with hydrocephalus and cerebral aneurysm treated successfully by combined approaches at one-stage.

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INTRODUCTION

The prevalence of pituitary tumors has been reported to be as high as 94 ± 19.3 cases per 100,000 population, which account for the 10-12% of all intracranial neoplasms^{1, 2}. However, the association between pituitary adenomas and hydrocephalus is rare, and only single case reports have been previously described. Furthermore, literature review did not reveal any case having the simultaneous association of pituitary adenoma with both hydrocephalus and intracranial aneurysm.

The microsurgical transsphenoidal approach is the most commonly used surgical approach for pituitary adenoma and is highly effective². However, the transsphenoidal approach for giant pituitary adenomas may be complicated by incomplete resection because of limited visualization and inability to maintain free circulation of cerebrospinal fluid. We recently treated a patient who had a pituitary adenoma simultaneously associated with obstructive hydrocephalus and intracranial aneurysm treated successfully by a combined transcranial and transventricular approach.

CASE REPORT

A 59-year-old man was admitted to the hospital with a 1-month history of headache, dizziness, and visual disturbance. Physical examination showed bitemporal hemianopsia. Funduscopic examination showed slight atrophy of the optic fundi. Magnetic resonance imaging (MRI) scan showed a tumor (4.0 x 1.8 x 1.5 cm) located mainly outside the sella turcica with

suprasellar extension. The right internal carotid artery was encased by the tumor, and there was hydrocephalus associated with tumor extension into the third ventricle (Figure 1). Endocrinologic tests were normal including levels of prolactin, growth hormone, thyroid hormone (free T3 and T4), thyroid stimulating hormone, follicle stimulating hormone, lactate dehydrogenase, and adrenocorticotropic hormone.

Surgical resection of the tumor was done with general anesthesia and a combined transcranial (subfrontal) and transventricular approach, with the patient positioned supine. The coronal incision was made just behind the hairline from the left to right side of the zygomatic arch. The craniotomy (length, 6 cm) was made 5 cm lateral to the midline; the frontal bone flap was isolated freely and the bottom of the craniotomy bone flap was made as low as possible. The dura was opened as a medial-based flap, and the large vein to the midline or sagittal sinus was preserved. A ventricular catheter (12 French) was initially inserted into the right lateral ventricle and third ventricle, and cerebrospinal fluid (30 mL) was drained to improve exposure. Then, a 3-cm incision was made in the right middle frontal gyrus to increase exposure. The tumor that had extended to the lateral ventricle and third ventricle was debulked, and the boundary between the tumor and surrounding structures was separated. The excision of residual tumor involving the internal carotid artery and intra-suprasellar region was performed from the right subfrontal approach. An incidental aneurysm (neck width, 6.0 mm) was noted in the right anterior

communicating artery, and the aneurysmal fundus projected directly to left anteroinferior direction (**Figure 1**). The anterior communicating artery was clipped because of the risk of bleeding from the aneurysm during the resection of the pituitary adenoma. The tiny vessels, especially the perforating arteries to the hypothalamus, were protected from injury. Histopathologic examination of the specimen showed a non-functioning pituitary adenoma.

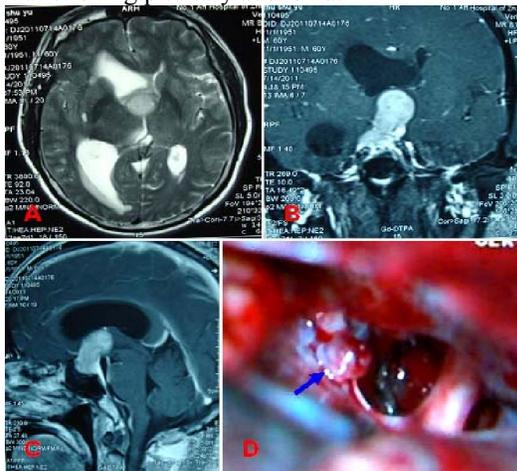


Figure 1.

After surgery, the patient had visual improvement and no major complications except for transient diabetes insipidus on the third postoperative day, which resolved quickly after administration of desmopressin. He did not develop postoperative panhypopituitarism, vasospasm, or ischemic attack. Postoperative MRI scan at 3 months after microsurgery showed no residual tumor or hydrocephalus, and satisfactory clipping of the anterior communicating artery aneurysm was confirmed with a 64-slice CTA (**Figure 2**). The patient had gradual improvement of symptoms, and his hormonal status was normal through follow-up at 6 months after surgery.

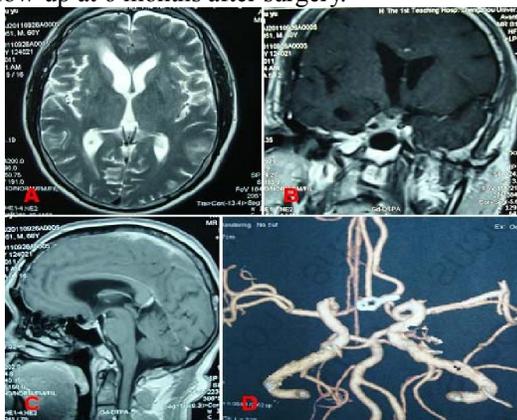


Figure 2.

DISCUSSION

The present case was unusual because the patient had a pituitary adenoma simultaneously

associated with both obstructive hydrocephalus and intracranial aneurysm. Previous cases of pituitary adenoma occasionally had been reported associated with either hydrocephalus (**Table 1**) or intracerebral aneurysm (**Table 2**), but giant pituitary adenoma with hydrocephalus and cerebral aneurysm simultaneously have not been reported based on previous literatures. The present patient had normal endocrinologic tests, consistent with a non-functioning pituitary tumor, and serendipitous discovery of the intracranial aneurysm, during surgery to resect the pituitary adenoma, complicated the treatment strategy.

Microsurgery frequently is used for treatment of non-functioning pituitary adenomas. Diverse surgical strategies for the treatment of pituitary adenoma with hydrocephalus have included as following: ventriculoperitoneal shunting before transcranial resection of the pituitary tumor³; external ventricular drainage and medical treatment^{4,5}; external ventricular drainage before definitive surgical intervention⁶; and combined transsphenoidal and transventricular approaches^{7,8} (**Table 1**). In the present case, a combined approach was used, including subfrontal (transcranial) and transventricular approach instead of ventriculoperitoneal shunting or external drainage, because the hydrocephalus usually resolved when the tumor was removed according to our clinical practice, in addition, the potential infection and catheter-related complications such as drainage failure were also avoided. Therefore, it is our primary goal to ensure cerebrospinal fluid flowing easily through tumor resection. And gross total resection and resolution of hydrocephalus were obtained in a 1-stage procedure in this patient.

The combined approach provided the best visualization of critical neurovascular structures during tumor resection^{9,10,11}. A simultaneous “above and below” technique has been described, which combines a transsphenoidal approach with a craniotomy; with this approach, 40% gross total resection was achieved in 10 patients, and many with preoperative visual field defects had complete improvement⁹. The combined, simultaneous transsphenoidal and pterional approach may be indicated for a small subset of patients with large nonfunctional pituitary tumors who have a tumor configuration that cannot be completely resected by a single approach⁶. In a previously reported patient with hydrocephalus caused by giant pituitary adenoma, the tumor was resected totally with combined transsphenoidal and simultaneous transventricular endoscopic decompression⁷. Usually, microsurgical treatment is recommended for most giant pituitary adenomas using a transsphenoidal approach because of minimal surgical morbidity. However, complete resection of tumors with excessive suprasellar extension may be difficult to achieve and the

procedures may be associated with major morbidity or mortality. Although endoscopy-assisted microsurgical treatment could provide satisfactory tumor resection with low risk of postoperative complications, the necessary specialized endoscopic equipment was not available in our hospital. Nevertheless, the subfrontal (transcranial) and transventricular approach was effective for treatment of hydrocephalus associated with the pituitary adenoma based on our previous experience.

A previous patient with a pituitary adenoma had died because of hemorrhage from a solitary subfrontal approach⁶ In that case, the hemorrhage had been difficult to manage because the tumor had extended into the hypothalamus and third ventricle and

there was a blind zone near the posterior suprasellar cistern and vital fenestrated vessel. Improved exposure with a combined approach may have helped to treat the hydrocephalus and pituitary adenoma with lower risk of hemorrhage. In present rare case, the combined approach enabled performance of the resection in 1 stage and provided excellent exposure. This improved exposure enabled early visualization and preservation of the pituitary stalk, and avoided potential hypothalamic injury from excessive retraction and inadequate hemostasis. Therefore, the combined approaches may achieve the goal of more tumor resection with less need for multiple sequential operations.

Table 1. Previously Reported Cases Identified with Pituitary Adenoma and Hydrocephalus*

Case number	Reference	Year	Age	Sex†	Symptoms	Endocrinologic tests	Surgical approach/treatment	Outcome	Complication
1	Verhelst [3]	1998	61	M	Disturbed vision Somnolence	Normal	VP shunt Transcranial Transsphenoidal	PR	Deteriorated vision (temporary)
2	Zikel [4]	1999	81	F	Decreased consciousness	Increased prolactin	Medical treatment	TS	None
3	Scarone [5]	2006	26	M	Headache Nausea Vomiting	Increased prolactin	EVD Medical treatment	TS	None
4	Joshi [6]	2009	72	M	Disturbed vision Confusion	NA	Transsphenoidal	PR	Hemorrhage Death
5	Joshi [6]	2009	71	M	Parkinson disease	NA	EVD Transsphenoidal	PR	Hemorrhage
6	Joshi [6]	2009	32	M	Headache Confusion Disturbed vision	NA	EVD Transsphenoidal	PR	Poor vision
7	Joshi [6]	2009	42	M	Altered personality Disturbed vision	NA	EVD Transsphenoidal	NA	Poor vision
8	Joshi [6]	2009	42	F	Decreased consciousness	Increased prolactin	EVD Transcranial	PR	None
9	Ojha [7]	2009	38	M	Increased intracranial pressure Visual disturbance Hypothalamic disturbance	Low cortisol	Transsphenoidal Transventricular	GTR	None
10	Romano [8]	2010	71	F	Headache Spatial disorientation Drowsiness Generalized weakness	Panhypopituitarism	Transsphenoidal Transventricular	GTR	None
11	Present case	2012	59	M	Headache Dizziness Visual disturbance	Normal	Transcranial Transventricular	GTR	None

*Abbreviations: EVD, external ventricular drainage; GTR, gross total removal; NA, not available; PR, partial removal; TS, tumor shrinkage, † Sex: M, male; F, female.

Table 2. Previously Reported Cases Identified with Pituitary Adenoma and Intracranial Aneurysm*

Case number	Reference	Year	Age	Sex†	Symptoms	Aneurysm location	Aneurysm surgery	Pituitary surgery/treatment	Outcome‡
1	Hardy [14]	1991	27	F	Headache	Right ACA	Clipping	Biopsy	Excellent
2	Hermier [15]	1994	58	M	Visual loss Asthenia	ACA	Clipping	Frontopterional	Excellent
3	Imamura [16]	1998	72	F	Epistaxis	ICA-cavernous	None	None	Died
4	Revuelta [17]	2002	60	F	Headache Visual loss	Supraclinoid ICA	Clipping	Supraorbital keyhole	Excellent
5	Sade [18]	2004	39	F	Acromegaly	ICA-cavernous	Embolization	Transsphenoidal	Excellent
6	Bulsara [19]	2007	73	M	Headache Blurred vision Photophobia	ACA	Clipping	Transcranial	Excellent
7	Seda [20]	2008	58	F	Headache Acromegaly	ICA-ophthalmic artery	Clipping	Transsphenoidal	Excellent
8	Soni [21]	2008	53	M	Headache Visual disturbance Ophthalmoplegia	ICA-cavernous	Embolization	Medical treatment	Excellent
9	Wang [22]	2009	61	F	Visual field defect	Right supraclinoid ICA	Embolization	Transsphenoidal §	Good
10	Nishijima [23]	2010	40	F	Headache Right hemiparesis	ICA-cavernous	Embolization	Transsphenoidal	Excellent
11	Present case	2012	59	M	Headache Dizziness Visual disturbance	Right ACA	Clipping	Transcranial Transventricular	Excellent

* Abbreviations: ACA, anterior communicating artery; ICA, internal carotid artery.

† Sex: M, male; F, female.

‡ Outcome: excellent: improved symptoms; good, no major changes compared with preoperative assessment.

§ Case 9 also had adjuvant treatment with X-knife radiotherapy; no other case had adjuvant treatment.

Intracranial aneurysm has been observed in 3.7% to 7.4% patients with pituitary adenoma^{12, 13}, primarily as solitary rare cases¹⁴⁻²³ (Table 2). The presence of an intracranial aneurysm together with a pituitary adenoma may be associated with high risk to the patient, especially when the aneurysm is near the operative field. Furthermore, the additional finding of hydrocephalus in this patient added to the challenge for suitable treatment because limited experience was available about adenomas with such complex associations. Therefore, the first step of microsurgical treatment was to remove the tumor extending into the lateral ventricle and third ventricle, to resolution of CSF obstruction. After completing the tumor resection from the lateral ventricle and third ventricle, the residual tumor involving the internal carotid artery and suprasellar region was removed by the subfrontal approach, and the incidental aneurysm was found in the anterior communicating artery. Disastrous bleeding could have occurred at any time during the resection of the pituitary adenoma, so aneurysm clipping was preferred, it must be noted that it is impossible to clip the aneurysm if surgical approach was adopted by transsphenoidal rather than the subfrontal approach in this case; subsequently, residual tumor involving the internal carotid artery and intra-suprasellar region was performed successfully by the right subfrontal approach.

This combined approach was effective treatment for the pituitary adenoma associated with hydrocephalus and cerebral aneurysm.

Conclusions

The co-existing of giant pituitary adenomas with hydrocephalus and cerebral aneurysm is extremely rare. To the best of our knowledge, this is the first report of giant pituitary adenomas with hydrocephalus and cerebral aneurysm treated successfully, surgical resection of giant pituitary adenomas with hydrocephalus and clipping cerebral aneurysm in one patient remain challenging lesions with high risk, the present study demonstrates that the combined subfrontal plus transventricular approach was an valuable and effective management for complex pituitary adenoma. The most striking advantage of combined approach could potentially improve gross total resection and solve the hydrocephalus as well as clip the accompanying aneurysm at one-stage.

Conflict of interest: None.

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