Pituitary apoplexy accompanying temporal lobe epilepsy as a complication: case report

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Case report

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Abstract

Abstract Background: Pituitary apoplexy is an acute clinical syndrome caused by infarction and/or hemorrhage of pituitary adenoma, which typically presents with severe headache, visual deterioration, and endocrine abnormalities. However, temporal lobe epileptic seizure has not been viewed as a symptom of pituitary apoplexy in the literature. Case presentation: To elucidate further such a rare complication of temporal lobe epilepsy, we describe here the rare clinical manifestations of a 55-year-old previously healthy male with pituitary apoplexy harboring headache, combined palsies involving cranial nerves III to VI, endocrinologic disturbances, and temporal lobe epilepsy. In addition, we discuss the temporal lobe epilepsy of pituitary adenoma based on the literature. Conclusion: Although further accumulation of clinical data is needed, we would like to emphasize the importance of recognition of temporal lobe epilepsy due to pituitary apoplexy, and suggest that early surgery should be considered as an option in patients with such a rare complication.

Background

Pituitary apoplexy is a clinical syndrome that is thought to be caused by infarction, hemorrhage, or hemorrhagic infarction within a preexisting pituitary adenoma [1-4, 8-14, 18-21, 25-28]. However, pituitary apoplexy is confounded by the inclusion, in some series, of pituitary adenomas with histopathologic or radiologic evidence of hemorrhage but without sudden onset of clinical symptoms [14, 28]. The classical term pituitary apoplexy presentation includes the abrupt onset of severe headache accompanied by neurologic, ophthalmologic, and/or endocrinologic deterioration due to sudden expansion of a mass within the sella turcica [1-4, 8-14, 18-21, 25-28]. It is a rare event that occurs in 0.6 - 17% of pituitary adenomas [1-4, 8-14, 18-21, 25-28]. The most common presenting symptom is headache with a frequency varying from 63 - 100% [1-4, 8-14, 18-21, 25-28]. Visual abnormalities such as visual field deficit, impaired visual acuity, and/or ocular palsies may be found in 50 - 80% of patients [1-4, 8-14, 18-21, 25-28], due to compression of the optic chiasm and cranial nerves in the cavernous sinus. Other symptoms, such as nausea and vomiting which may be a result of headache, meningeal irritation, intracranial hypertension, and/or acute adrenal insufficiency, are found in 25 - 80% of cases [1-4, 8-14, 18-21, 25-28]. However, according to our understanding, temporal lobe epilepsy is not viewed as a symptom of pituitary apoplexy in the literature [1-4, 8-14, 18-21, 25-28].

To elucidate such a rare complication of temporal lobe epilepsy further, we describe here the rare clinical manifestations of a 55-year-old male with pituitary apoplexy harboring headache, ocular palsies, hypoesthesia in the second division of the trigeminal nerve, endocrinologic disturbances, and temporal lobe epilepsy. In addition, we present a small literature review of temporal lobe epilepsy elated to pituitary adenoma.

Case Presentation

A 55-year-old male was referred to our department, because computed tomography (CT) and magnetic resonance imaging (MRI) of the head undertaken at another hospital had disclosed an intra- and suprasellar abnormality. Three days prior to the CT and MRI, he had experienced slight left eye ptosis, accompanied by diplopia and retrobulbar pain. The following morning, he developed unusual headache. Prior to this event, he had been in good health. At admission, his consciousness level was clear but he demonstrated severe headache, left diplopia, eyelid ptosis, and facial numbness. On neurologic examination, restriction of lateral, medial, downward, and upward movement of the left eye was observed, and his left pupil was enlarged (approximately 5 mm) and unresponsive to light. Furthermore, he experienced hypesthesia near his left cheek. However, there was no impairment of his visual acuity, and no hemianopsia. These observations indicated left oculomotor nerve, trochlear nerve, abducens nerve, and trigeminal nerve (maxillary nerve) palsies. Laboratory blood tests revealed the following: WBC count, 10,300/mm3; Hb, 14.2 g/dL; Ht, 40.7%; PLT count, 228,000/mm3; CRP, 8.62; Na, 133 mmol/L; K, 4.6 mmol/L; and Cl, 94 mmol/L. His plasma hormone levels were as follows: prolactin, 6.59 ng/mL (normal range, 3.6 - 16.3 ng/mL); growth hormone, 1.70 ng/mL (- 2.47 ng/mL); insulin-like growth factor-1, 219 ng/mL (84 - 238 ng/mL); adrenocorticotropic hormone (ACTH), 2.7 pg/mL (7.2 - 63.3 pg/mL); cortisol, 1.42 mg/dL (6.24 - 18 ug/dL); thyroid-stimulating hormone, 0.23 uIU/mL (0.34 – 3.8 uIU/mL); free T3, 2.71 pg/mL (2.0 - 3.8 pg/mL); free T4, 1.38 ng/dL (0.8 - 1.5 ng/dL); luteinizing hormone, 0.8 mIU/mL (1.8–5.2 mIU/mL); and follicle-stimulating hormone, 2.2 mIU/mL (2.9–8.2 mIU/mL). These findings were almost consistent with ACTH deficiency (subsequently with reduction of corticotropin release) and gonadotropin deficiency. Replacement hormone therapy was therefore initiated with hydrocortisone.

CT and MRI of the head displayed a nonhomogenous sellar and suprasellar expansile lesion which was attached to the optic chiasm indicative of pituitary apoplexy. Furthermore, this sellar mass lesion revealed significant extension into the left cavernous sinus and compression of the mesial temporal lobe, but there was absence of signs of subarachnoid hemorrhage. Diffusion-weighted and T2-weighted MRI demonstrated a markedly increased signal intensity area within the pituitary mass. Gadolinium enhanced MRI showed peripheral enhancement surrounding the pituitary mass (Figs. 1 and Fig. 2).

During informed consent, he suddenly displayed prominent motor arrest with loss of awareness and commenced oroalimentary automatism (chewing) and gestural automatism (repetitive motor action of the right upper limb). A few minutes later, he began to experience a generalized convulsion. This was treated with intravenous administered diazepam. After this event, antiepileptic treatment (levetiracetam) was initiated.

On the day of admission to our institution, endoscopic transnasal transsphenoidal surgery was performed to remove the mass, which was soft and easy to curette. During the surgery, a low level of spinal fluid leakage was noted, but no subarachnoid hemorrhage was observed. Microscopic examinations of the surgical specimen demonstrated a large area that consisted primarily of coagulative necrosis and hemorrhage. A typical adenoma pattern forming diffuse sheets was still evident in the tumor tissues (Fig. 3). These findings were consistent with pituitary apoplexy.

The patient was discharged from our hospital 20 days after surgery, and his postoperative course was uneventful. MRI at the time of discharge demonstrated gross total removal of the pituitary mass and no evidence of residual tumor. The diminished mass effect of the pituitary lesion resulted in improvement of the compression to the optic chiasm and mesial temporal lobe. Furthermore, MR images did not show edema and hemosiderin in the perilesional area of the temporal lobe (Fig. 4). His left oculomotor nerve deficits including eye ptosis and pupil dilatation and facial hypesthesia due to trigeminal nerve deficit gradually improved, although his abducens nerve palsy persisted after the surgery. An electroencephalogram at 20 days following surgery showed no epileptiform activity. The patient is receiving corticosteroid replacement therapy and anticonvulsant medication, and continues to be followed up.

Discussion And Conclusions

Even though several pathomechanisms have been suggested to underlie the development of spontaneous pituitary apoplexy, the true pathogenesis remains unclear [1-4, 8-14, 18-21, 25-28]. Presumably, each mechanism could contribute to the acute deterioration of an already compromised blood supply to the pituitary tumor [4, 28]. Most cases develop spontaneously, however, several reports including our past paper (28), have suggested various conditions, procedures, and treatments could cause pituitary apoplexy; such as diabetes mellitus, head trauma, hypertension, pregnancy, carotid angiography, lumbar puncture, myelography, pituitary function tests, pneumoencephalography, anticoagulant therapy, bromocriptine therapy, and radiotherapy [1-5, 8-14, 18-21, 25-28]. Typical pituitary apoplexy presents severe headache, loss of vision, and endocrine abnormalities. However, we have been unable to uncover any previous reports of temporal lobe epileptic seizure occurring as a complication of pituitary apoplexy [1-4, 8-14, 18-21, 25-28].

It is easy to speculate that the pituitary adenoma, which compresses or infiltrates the inner structure of the temporal lobe due to lateral extension, can induce temporal lobe epileptic seizures. Previously, Elkington in 1986 reported that 6 cases out of 260 pituitary adenomas (2.3%) developed epileptic seizure as a preoperative symptom; details were uncertain, but 4 of the 6 cases were grand mal, one was temporal lobe epilepsy, and one was uncinate fits [6]. Deepak et al. described that a lot of information about pituitary adenoma related seizures including exact prevalence, their clinical and electrophysiological characteristics, and treatment is inadequate [5]. Even now, such details have not been fully elucidated.

On the other hand, epileptic seizures have been observed as a complication of medically treated prolactin-producing macroadenoma. Niwa et al. (Japanese-language paper) reported that treatment with bromocriptine alone for prolactin-producing macroadenomas with lateral extension to the cavernous sinus revealed symptomatic seizures during the medical course in 3 out of 9 cases, although in all cases a rapid decrease of serum prolactin level was recognized and in 8 cases evidence of tumor shrinkage was found on CT and/or MRI. Additionally, a hypointense signal on both T1- and T2-weighted images, consistent with hemosiderin due to intratumoral hemorrhage caused by bromocriptine, was recognized

around the residual tumor in 3 cases with seizure. They suggested therefore that hemosiderin deposition within the medial surface of the temporal lobe, following bromocriptine therapy for prolactin-producing macroadenomas with intradural supracavernous extension, could be a trigger of such seizures [17]. Thus, if the hemosiderin remains within the adenoma, no seizures will occur, but if the hemosiderin escapes from the adenoma and comes into contact with the medial aspect of the temporal lobe, it could trigger a seizure attack. Deepak et al. reported that 6 out of 29 patients (5 cases of temporal lobe epilepsy and one presenting with grand mal seizure), diagnosed as having macroprolactinoma, which invade the brain parenchyma, had a history of epilepsy prior to or at presentation of pituitary adenomas. In contrast, the remaining 23 patients showed no clinical symptoms suggesting epileptic activity despite the presence of brain infiltration by the adenoma on neuroimaging. The median serum prolactin level of those with epilepsy was 369,000 mU/l. All 5 patients with temporal epilepsy experienced an immediate reduction in the occurrence of seizures following initiation of dopamine agonist treatment, which coincided with the fall in their serum prolactin levels but with minimal or no radiologic change in the appearance of their tumors, and additionally it was possible to reduce their dose of anti-epileptic medication. It was suggested that dopamine agonist therapy, which has an intrinsic inhibitory anti-epileptogenic function, can reduce the ictal frequency and doses of anti-epileptic drugs [5]. On the other hand, gamma knife surgery can serve as a therapeutic tool for uncontrolled pituitary adenoma and for mesial temporal lobe epilepsy, but radiation induced MR changes outside of the radiosurgical target and associated transient increases of seizure rate are well-known side effects of radiosurgery [22]. Schindler et al. reported the first case of radiation-induced changes to the mesial temporal lobe structures and transient symptomatic epileptic seizures as rare side-effects of gamma knife radiosurgery of the cavernous sinus in residual growth hormone secreting pituitary adenoma. The MR changes of their patient were temporary and diffuse hyperintensity lesion of T2-weighted image with central enhancement was noted in the left medial temporal lobe [22]. Follow-up with future imaging is necessary in our case: compression of the internal structure of the temporal lobe could trigger temporal epileptic seizures since there have been no changes in the temporal lobe images. In particular, our case indicated lateral infiltration: Group 4 on the Knosp classification was estimated from the image at onset, and unusual combined palsies involving cranial nerves III to VI were observed [15], which was remarkable as compared with the upper part of the sellar. It is inferred that such unusual compression elicited temporal lobe epileptic seizures.

There is as yet no clear consensus regarding the best option for the management of pituitary apoplexy [1-4, 7-14, 18-21, 24-29]. However, classical pituitary apoplexy represents one of the major medical emergencies, and there is no doubt that immediate fluid administration and replacement with hydrocortisone to achieve hemodynamic stability and to reduce hormone deficiencies are important for saving lives [1-4, 7-14, 18-21, 25-28]. On the other hand, the role of pituitary surgery and the timing of surgical decompression of the pituitary fossa remain topics of debate [1-4, 7-14, 18-21, 25-29]. Hitherto, it has been widely accepted that transsphenoidal surgery can be minimally invasive within the first week and should be considered in the presence of severe visual loss for pituitary apoplexy, and that patients without severe visual loss or consciousness impairment may be treated expectantly by conservative management [1-4, 7-14, 18-21, 24-29]. Temporal lobe epilepsy induced by pituitary apoplexy is a rare

symptom, and it has not been viewed as a complication in the literature [1-4, 8-14, 18-21, 25-28]. However, recognition and optimal treatment of temporal lobe epilepsy as complication of pituitary apoplexy are important, because patients with epilepsy exhibit a diminished quality of life and a high mortality rate as compared to the general population [22, 23], and patients with temporal lobe epilepsy are frequently resistant to anti-epileptic medication [16, 23]. Furthermore, involvement of epileptogenicity changes over the course of time should be eliminated, although the true pathogenesis of temporal lobe epilepsy as a complication of pituitary apoplexy still remains unclear. Therefore, although further accumulation of clinical data is needed, we would like to suggest that early surgery should be considered as an option in patients with temporal lobe epilepsy occurring as a complication of pituitary apoplexy.

Abbreviations

CT: computed tomography; MRI: magnetic resonance imaging; WBC: white blood cell; Hb: hemoglobin; Ht: hematocrit; PLT: platelet; CRP: carbon reactive protein; Na: sodium; K: potassium; Cl: chorine; ACTH: adrenocorticotropic hormone.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

The patient and next of kin have consented to submission of the case report to the journal, and we have obtained a written informed consent.

Availability of data and materials

All data related to this case report are contained within the manuscript.

Competing interests

All authors have no affiliations with or involvement in any organization or entity with any financial interest, or non-financial interest in the subject matter or materials discussed in this case report.

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Authors' contributions

KS and AY contributed the concept of the manuscript and drafted the manuscript. HO and SY revised the manuscript and contributed the conception of the manuscript. SY, FM, and KS contributed to the

obtaining and interpreting of the clinical information. All authors read and approved the final version of the manuscript.

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Figures

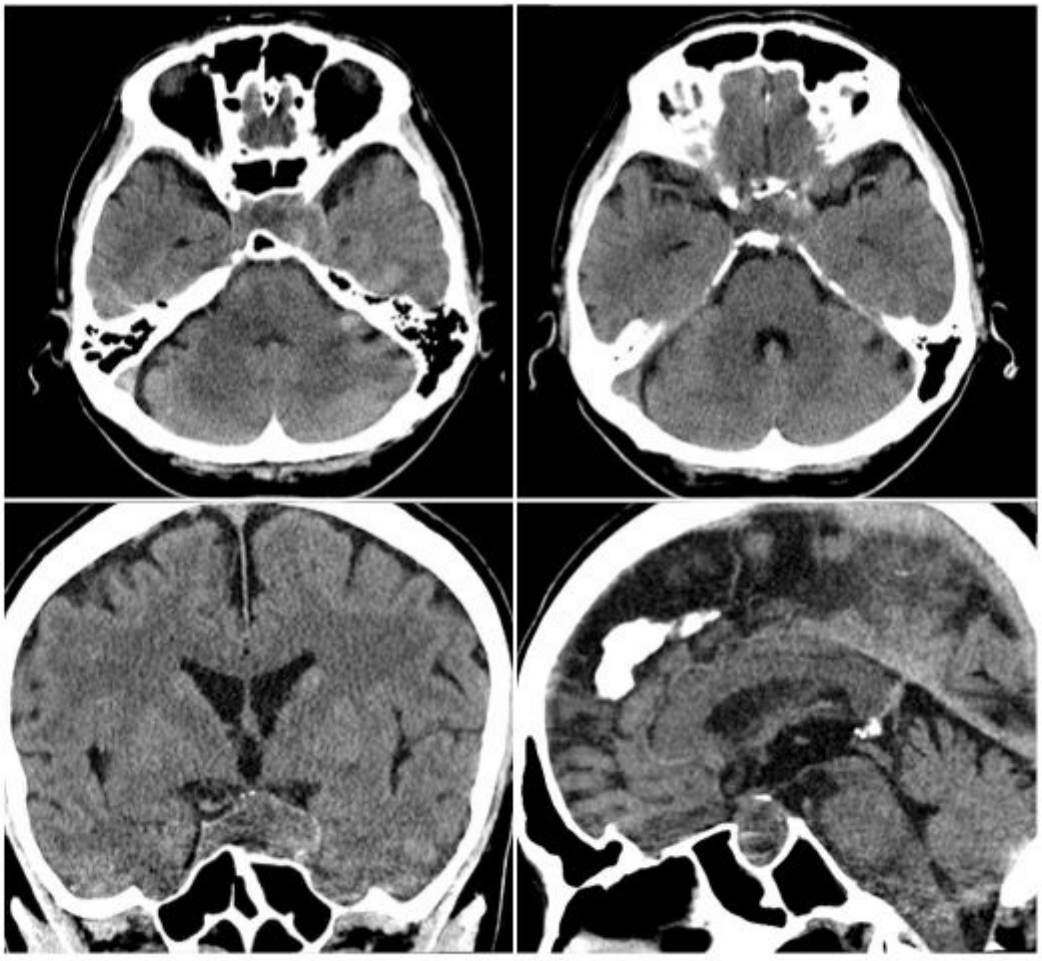

Figure 1

Plain computed tomography (CT) of the head disclosing an enlarged sellar mass lesion with a partially slight high density, but no evidence of subarachnoid hemorrhage

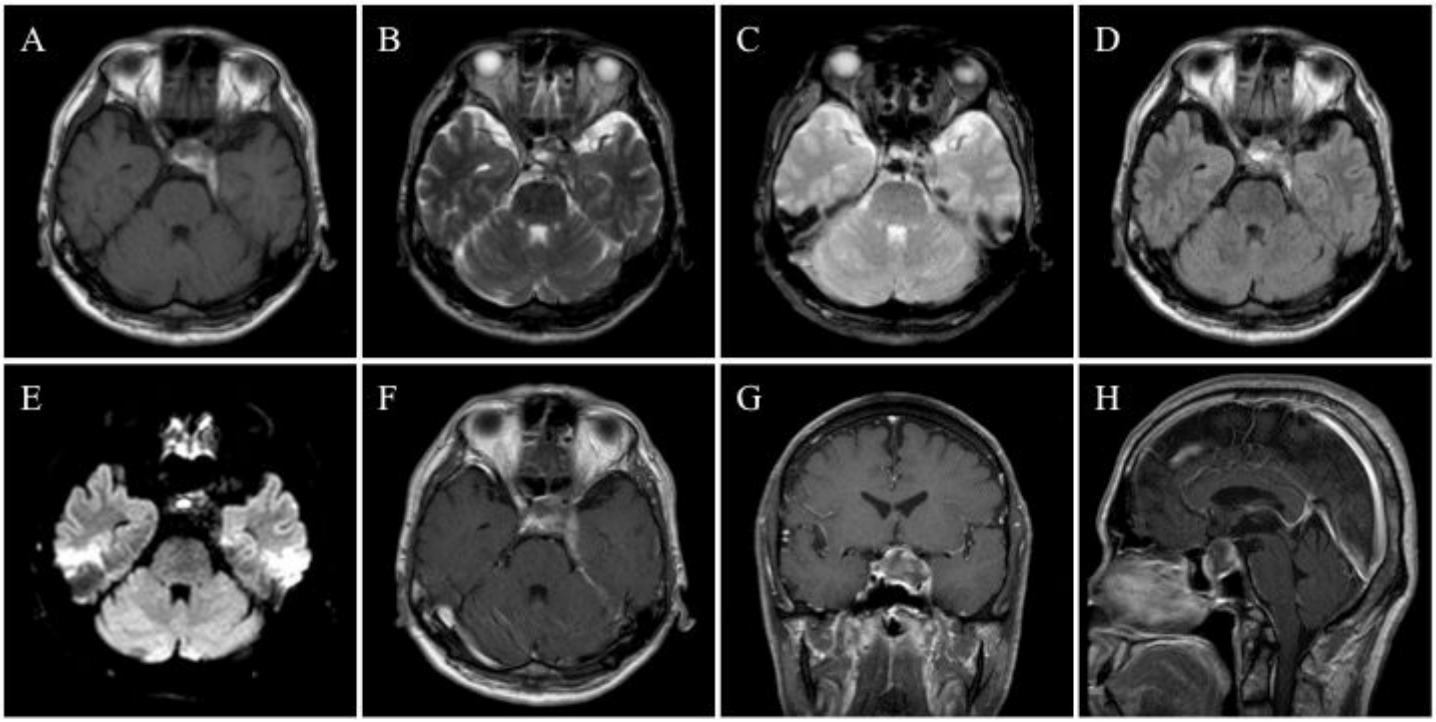

Figure 2

Magnetic resonance imaging (MRI) revealing a nonhomogenous mass filling the sella with extension into the suprasellar region and entering the left cavernous sinus. The images show axial T1-weighted (A), T2-weighted (B), T2*-weighted (C), fluid-attenuated inversion recovery (FLAIR) (D), and diffusion-weighted (E) MRI. Furthermore, gadolinium enhanced T1-weighted axial (F), coronal (G), and sagittal (H) MRI demonstrated peripheral enhancement surrounding the pituitary mass lesion, attachment to the optic chiasm, and a compressed mesial temporal lobe.

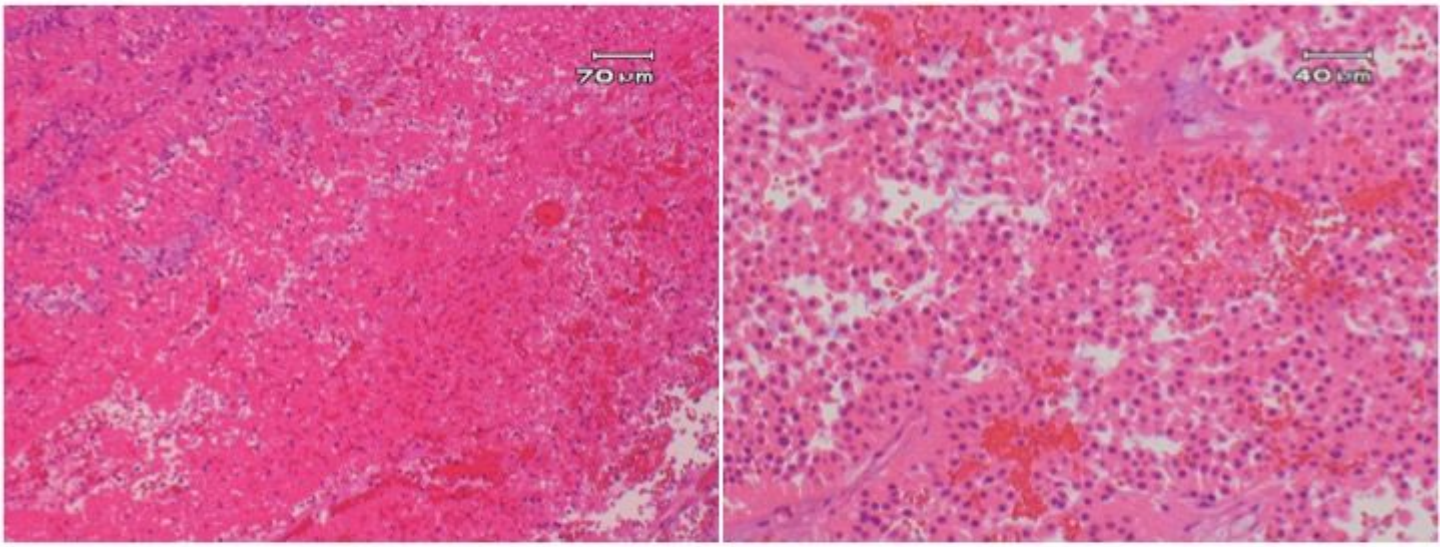

Figure 3

Histological examinations of the surgical specimen revealed large areas that had recently been almost totally infarcted (left: low magnification image, hematoxylin and eosin staining). A typical adenoma pattern, which was eosinophilic, of high cellularity, and formed diffuse sheets, was still demonstrated in the residual tumor tissues (right: high magnification image).

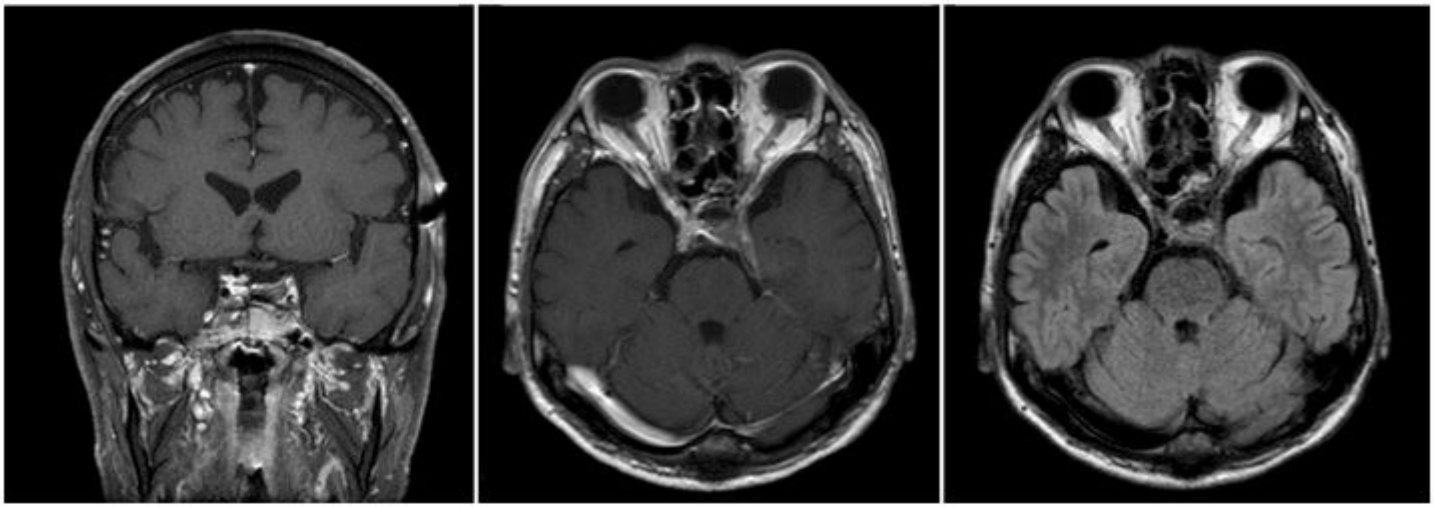

Figure 4

Immediate post-operative MRI demonstrating removal of the sellar mass including part of left cavernous sinus. Gadolinium enhanced T1-weighted coronal (A), axial (A), and FLAIR (C) MRI showed no attachment to the optic chiasm and no compression of the mesial temporal lobe.

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