Ruptured aneurysm–induced pituitary apoplexy: illustrative case

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BACKGROUND Pituitary apoplexy associated with aneurysmal rupture is extremely rare and may be misdiagnosed as primary pituitary adenoma apoplexy. The authors present a case of a patient with pituitary apoplexy caused by rupture of an anterior cerebral artery aneurysm embedded within a giant pituitary adenoma, and they review the relevant literature.

OBSERVATIONS A 78-year-old man experienced sudden headache with progressive vision loss. Magnetic resonance imaging (MRI) revealed a giant pituitary tumor with abnormal signal intensity. Magnetic resonance angiography immediately before surgery showed a right A1 segment aneurysm, suggesting coexisting pituitary apoplexy and ruptured aneurysm. The patient underwent urgent transsphenoidal surgery for pituitary apoplexy. The tumor was partially removed, but the perianeurysmal component was left behind. Subsequent cerebral angiography showed a 5-mm right A1 aneurysm with a bleb that was successfully embolized with coils. Retrospective review of preoperative dynamic MRI showed extravasation of contrast medium from the ruptured aneurysm into the pituitary adenoma. Histopathologic examination showed gonadotroph adenoma with hemorrhagic necrosis. Postoperatively, the patient’s visual function improved.

LESSONS MRI identification of pituitary apoplexy caused by aneurysmal rupture has not been reported previously. Aneurysmal rupture should be considered in the differential diagnosis of pituitary apoplexy. When a ruptured aneurysm is encountered, the authors recommend treating it before addressing pituitary apoplexy.

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KEYWORDS anterior cerebral artery aneurysm; nonfunctioning pituitary adenoma; pituitary apoplexy; ruptured aneurysm

Pituitary apoplexy is a clinical syndrome caused by acute hemorrhage or ischemic infarction of the pituitary gland. Pituitary apoplexy associated with a ruptured aneurysm is extremely rare. 3 We report the first case of pituitary apoplexy caused by rupture of an A1 segment anterior cerebral artery aneurysm that was embedded within a giant nonfunctioning pituitary adenoma, and we review the pathophysiology and relevant literature.

Illustrative Case

A 78-year-old right-handed man experienced sudden headache and nausea with bitemporal hemianopia and collapsed. On admission to the nearest emergency hospital, brain computed tomography (CT) revealed a high-density mass arising from the sella. Magnetic resonance imaging (MRI) performed 1 day later showed pituitary apoplexy with subarachnoid hemorrhage (SAH).

Three days after symptom onset, the patient was transferred to our hospital because of progressive vision impairment. Neurological examination showed disorientation, right ptosis, bilateral unresponsive pupils, and absence of extraocular movements in all directions. His left eye was blind. In the right eye, light perception was poorly preserved. The finding of endocrinological testing was within normal range.

Repeat imaging studies showed a 5-cm × 4-cm × 3-cm pituitary macroadenoma with suprasellar extension and right cavernous sinus invasion (Fig. 1A and B). Diffusion-weighted sequences showed high signal within the intrasellar mass. Magnetic resonance angiography immediately before surgery showed a right A1 aneurysm embedded within the tumor. Hematoma was distributed mainly around the aneurysm, suggesting coexisting pituitary apoplexy and ruptured aneurysm. Urgent endoscopic transsphenoidal surgery was performed. The tumor was

ABBREVIATIONS CT = computed tomography; GH = growth hormone; MRI = magnetic resonance imaging; PRL = prolactin; SAH = subarachnoid hemorrhage.

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partially removed, and the perianeurysmal component was intentionally left behind. Cerebral angiography on the morning after surgery confirmed an irregular 5-mm right A1 aneurysm with a superoposteriorly projecting bleb, which was the presumed rupture point (Fig. 1C and D, and 2A). Complete endovascular occlusion of the aneurysm was then achieved using platinum coils (Fig. 2B). Retrospective review of the preoperative dynamic MRI showed extravasation of contrast medium from the ruptured aneurysm into the pituitary adenoma (Video 1).

VIDEO 1. Clip showing preoperative dynamic MRI depicting extravasation of contrast medium from the ruptured A1 aneurysm into the pituitary adenoma. Click here to view.

Postoperative MRI showed partial tumor removal as expected; the perianeurysmal component remained. Histopathological examination of the surgical tumor specimen revealed both hemorrhagic necrosis (Fig. 3) and typical adenoma on hematoxylin and eosin staining; a typical adenoma was observed over a relatively large area. Immunohistochemical staining was positive for lutetinizing hormone and follicle-stimulating hormone, consistent with gonadotroph adenoma. The patient’s visual acuity improved to light perception in the right eye and 0.05 in the left.

Discussion

Observations

Coexisting intracranial aneurysms are found in 2.3% to 7.4% of patients with pituitary adenoma,2–4 a higher prevalence than that in the general population and in patients harboring other brain tumors. However, pituitary apoplexy associated with intracranial aneurysmal rupture is extremely rare. To our knowledge, only 5 cases have been reported (Table 1).1,5–7 The prevalence of coexisting unruptured aneurysm is high in growth hormone (GH)-type pituitary adenomas, ranging from 2.3% to 13.8%. Conversely, the prevalence is low in prolactin (PRL)-type adenomas (0.9%–2.4%),2,3,8,9 which are occasionally aggressive and invasive.10 Two of the 5 reported patients with intracranial ruptured aneurysm associated with pituitary apoplexy harbored PRL-type adenomas, and one harbored a GH-type adenoma; only our case had a nonfunctioning adenoma.

Local circulatory stress, endocrinological effects, mechanical effects, and direct invasion have been proposed as mechanisms underlying aneurysm formation in patients with pituitary adenomas.2,11–13 However, the contribution of the pituitary adenoma is unclear.2,3 Mechanisms other than endocrinological effects may have been involved in our patient with nonfunctioning adenoma.

In a review of cerebral angiograms in 467 pituitary adenoma patients, Pant et al. found aneurysms in 25 (5.4%), 97% of which were located in the anterior circulation.3 Among the 5 cases of intracranial ruptured aneurysm associated with pituitary apoplexy
TABLE 1. Patients with pituitary apoplexy associated with ruptured intracranial aneurysm

| Authors | Case | Year | Sex | Location | Aneurysm Location | Location of Rupture | Extension | Clinical Manifestations | Treatment | Outcome | Pathological Diagnosis | Wall Invasion | Size | Pathological Diagnosis | Side of Aneurysm | Location | Location of Tumor |
|---------|------|------|-----|----------|------------------|---------------------|----------|------------------------|-----------|---------|-----------------------|-------------|------|----------------------|----------------|----------|-------------------|
| Laidlaw et al., 2003 | 1 | 2003 | 50/F | Lt A1 | 3 mm | Yes (rt) | Yes | Headache, vomiting, & visual impairment | Removal of pituitary tumor & clipping of Lt ruptured A1 aneurysm (open surgery) | GR | Micro GH secreting w/ hemorrhagic necrosis | Yes | 3 mm | Yes (rt) | Yes | Headache, vomiting, & visual impairment | Removal of pituitary tumor & clipping of Lt ruptured A1 aneurysm (open surgery) | GR | Micro GH secreting w/ hemorrhagic necrosis |
| Shahlaie et al., 2006 | 2 | 2006 | 46/F | Acom | 5 mm | No | No | Headache, nausea, & visual impairment | Clipping; conservative therapy for pituitary tumor | GR | Micro Hemorrhagic necrosis adenoma (no hormone data) | No | 5 mm | No | No | Headache, nausea, & visual impairment | Clipping; conservative therapy for pituitary tumor | GR | Micro Hemorrhagic necrosis adenoma (no hormone data) |
| Song et al., 2014 | 3 | 2014 | 2014 | Lt Pcom | 6 mm | No | No | Headache, vomiting, & blurred vision | Clipping & partial removal of pituitary tumor (open surgery); clipping & partial removal of pituitary tumor & clipping of Lt ruptured A1 aneurysm (open surgery) | GR | Micro Hemorrhagic necrosis adenoma | Yes (bilat) | 6 mm | No | No | Headache, vomiting, & blurred vision | Clipping & partial removal of pituitary tumor (open surgery); clipping & partial removal of pituitary tumor & clipping of Lt ruptured A1 aneurysm (open surgery) | GR | Micro Hemorrhagic necrosis adenoma |
| Xu et al., 2015 | 4 | 2015 | 49/M | Lt A1 | 5 mm | Yes (rt) | Yes | Headache, vomiting, & visual impairment | Clipping; conservative therapy for pituitary tumor; radiotherapy for residual tumor | MD | Giant Prolactinoma | Yes | 5 mm | Yes (rt) | Yes | Headache, vomiting, & visual impairment | Clipping; conservative therapy for pituitary tumor; radiotherapy for residual tumor | MD | Giant Prolactinoma |
| Yoshida et al., 2021 | 5 | 2021 | 78/F | Rt A1 | 5 mm | Yes (rt) | Yes | Headache, nausea, & progressive visual loss | Clipping & partial removal of pituitary tumor & clipping of Lt ruptured A1 aneurysm (open surgery); coiling | GR | Giant Gonadotroph adenoma w/ hemorrhagic necrosis | Yes | 5 mm | Yes (rt) | Yes | Headache, nausea, & progressive visual loss | Clipping & partial removal of pituitary tumor & clipping of Lt ruptured A1 aneurysm (open surgery); coiling | GR | Giant Gonadotroph adenoma w/ hemorrhagic necrosis |

A1 = A1 segment of the anterior cerebral artery; Acom = anterior communicating artery; GR = good recovery; MD = moderately disabled; Pcom = posterior communicating artery.

According to Oh et al., older age and cavernous sinus invasion correlate with increased incidence of intracranial aneurysm in patients with pituitary adenomas.2 Tumor growth causes microanatomical alterations in the cerebral circulation, blood flow modification, and greater hemodynamic stress, which predispose patients to aneurysm formation.3,11,14 In our 78-year-old patient, the pituitary tumor invaded the right cavernous sinus, and the right A1 aneurysm was embedded within the tumor. As the tumor gradually grew out of the sella and extended beyond the optic chiasm, it may have invaded and weakened the aneurysmal wall, causing rupture.

To the best of our knowledge, MRI identification of pituitary apoplexy caused by rupture of an intracranial aneurysm has not been reported previously. Increased intratumoral pressure at the time of aneurysmal rupture might have caused compression of the pituitary vessels, resulting in secondary pituitary apoplexy.

We performed endoscopic transsphenoidal surgery first because we detected the aneurysm immediately before surgery. Fortunately, our patient experienced improved visual function and avoided fatal aneurysmal rerupture. However, his aneurysm ruptured within the tumor. In general, aneurysm treatment should be given priority when aneurysmal SAH is suspected.

Lessons
MIRI identification of pituitary apoplexy caused by rupture of an intracranial aneurysm has not been reported previously. Aneurysmal rupture should be considered in the differential diagnosis of pituitary apoplexy. When encountered, we recommend treating the ruptured aneurysm before addressing pituitary apoplexy.

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Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Conception and design: Hiu, Yoshida, Matsuo. Acquisition of data: Hiu, Yoshida, Morikawa, Ujifuku. Analysis and interpretation of data: Hiu, Baba, Morikawa, Anda. Drafting the article: Hiu, Yoshida, Horie. Critically revising the article: Hiu, Yoshida, Baba, Morikawa, Horie, Ujifuku, Yoshida, Matsunaga, Niino, Xie, Izumo. Reviewed submitted version of manuscript: Hiu, Baba, Horie, Ujifuku, Niino. Approved the final version of the manuscript on behalf of all authors: Hiu.
Administrative/technical/material support: Morikawa. Study supervision: Hiu.

Supplemental Information
Videos
Video 1. https://vimeo.com/534462897.

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