Pituitary Apoplexy Manifesting as Massive Intracerebral Hemorrhage
—Case Report—

Kota KURISU,1 Masahito KAWABORI,1 Yoshimasa NIIYA,1 Yuzuru OHTA,1 Shoji MABUCHI,1 and Kiyohiro HOUKIN2

1Department of Neurosurgery, Otaru Municipal Medical Center, Otaru, Hokkaido; 2Department of Neurosurgery, Hokkaido University Graduate School of Medicine, Sapporo, Hokkaido

Abstract
A 68-year-old man presented with severe consciousness disturbance caused by pituitary apoplexy resulting in massive intracerebral hemorrhage (ICH). He had been periodically followed up for asymptomatic pituitary adenoma at another hospital for 8 years. Neuroimaging examination revealed pituitary apoplexy and massive ICH located in the left frontal lobe, and the ICH was directly connected to the intratumoral hemorrhage. The diagnosis was massive ICH from pituitary apoplexy. The patient underwent emergent evacuation of hematoma and removal of the pituitary adenoma via bi-frontal craniotomy. Postoperatively, he continued to exhibit deep consciousness disturbance and died 1 month after the operation. Pituitary apoplexy is usually characterized by intra-tumoral hemorrhage. The treatment strategy for asymptomatic pituitary adenoma is still controversial. This case shows that we should always consider the risk of pituitary apoplexy manifesting as ICH which may cause a fatal outcome.

Key words: fatal pituitary apoplexy, pituitary adenoma, intracerebral hemorrhage, emergent surgery

Introduction
Pituitary apoplexy is a syndrome caused by an acute ischemic or hemorrhagic event in a pituitary adenoma.16–18 Various precipitating events have been implicated in the pathogenesis of pituitary apoplexy such as head trauma, arterial hypertension, diabetes mellitus, pregnancy, estrogen or bromocriptine therapy, cardiac surgery, increased intracranial pressure, radiotherapy, dynamic pituitary function tests, or anticoagulation. However, the condition occurs spontaneously in the majority of patients with pituitary adenoma.16,20,24 The incidence of pituitary apoplexy in patients with pituitary adenoma is 0.6–23%.2,3,8,9,13,16,24 Pituitary apoplexy is characterized by sudden onset of headache, visual impairment, or ophthalmoplegia caused by acute enlargement of the tumor.3,6,11,26,27,30 Intra-tumoral hemorrhage is the most common type of hemorrhage, but subarachnoid hemorrhage (SAH),16,17,19,27 and intra-ventricular hemorrhage5,21,25 may also result from pituitary apoplexy. Only one case of intracerebral hemorrhage (ICH) has been associated with pituitary apoplexy.10

Here, we report a case of pituitary apoplexy with massive ICH which resulted in unfavorable outcome.
Fig. 1 Coronal T1-weighted magnetic resonance images with gadolinium administration performed June 2005 (A), August 2008 (B), and February 2011 (C) showing no enlargement of the intra- and supra-sellar tumor which was considered to be pituitary adenoma.

Fig. 2 Preoperative computed tomography scans showing massive intracerebral hemorrhage in the left frontal lobe and bilateral lateral ventricle hemorrhages, as well as the high density intra- and supra-sellar mass (arrow).

35.1 pg/ml, range 7.2–63.3.

Emergent computed tomography (CT) demonstrated a massive hematoma in the left frontal lobe and hematomas in the bilateral ventricles (Fig. 2), probably caused by intraventricular rupture of ICH. In addition, the intra- and supra-sellar high density mass was also observed. Three-dimensional CT angiography was performed to exclude vascular abnormalities, such as aneurysm or arteriovenous malformation. Only elevation of A1 segment in both anterior cerebral arteries was detected and no vascular abnormalities potentially causing the massive ICH were identified. MR imaging revealed a large well enhanced mass located in the intra- and supra-sellar region indicating pituitary adenoma surrounded by and connecting to the massive ICH (Fig. 3).

Under a diagnosis of ICH caused by pituitary apoplexy, emergent surgery on the day of admission was carried out via bifrontal craniotomy. Preoperatively, replacement of hydrocortisone was performed. The ICH located in the left frontal lobe was evacuated after the corticotomy. After the ICH was totally evacuated, we confirmed that the hematoma was connected to the left lateral ventricle. Next, tumor resection was performed through the anterior interhemispheric approach. The white aspirable tumor was located in the sella turcica and extended to the supra-sellar region. The bilateral optic nerves and chiasma were markedly stretched and displaced superiorly. The right oculomotor nerve was also compressed by the tumor laterally. During resection of the tumor, we confirmed that the intra-tumoral hemorrhage connected to the ICH located in the left frontal lobe. These operative findings indicated that the left frontal ICH was formed as a result of pituitary apoplexy. The tumor was semi-totally removed except for the intra-sellar lesion, because we considered that the intra-sellar lesion could be removed more easily by trans-sphenoidal surgery in the chronic stage. During the resection, the pituitary stalk was preserved. Finally the ICH and supra-sellar tumor were totally removed.

Histological examination of the surgical samples revealed hemorrhagic changes and necrotic changes (Fig. 4) which may have lead to pituitary apoplexy. The histological diagnosis was pituitary apoplexy as a result of pituitary adenoma. Further immunohistological staining showed positive reaction for chromogranin, but not for prolactin, GH, ACTH, luteinizing hormone (LH), or follicle-stimulating hormone (FSH).

After the operation, he continued to exhibit deep consciousness disturbance. MR imaging revealed that mas-
sive ICH and the supra-sellar part of the pituitary adenoma had been totally removed (Fig. 5). Although we observed intraoperatively that the pituitary stalk and pituitary gland were preserved intact, he developed panhypopituitarism (PRL 1.02 ng/ml, GH 0.22 ng/ml, ACTH <0.10 μIU/ml, FSH 0.30 mIU/ml, TSH <0.005 μIU/ml). However, postoperative diabetes insipidus did not occur. Administration of hydrocortisone and thyroid hormone replacement for hormonal therapy was performed. However, his general condition gradually worsened. He suffered severe aspiration pneumonia. Antibiotic medication was not effective. Unfortunately he died 1 month after the operation of progression of multiple organ failure.

Discussion

Pituitary apoplexy usually manifests as acute symptoms which may include headache, pituitary insufficiency, impaired consciousness, visual worsening and/or ophthalmoplegia. Acute tumor enlargement caused by intratumoral hemorrhage and compression of the surrounding tissues is important in causing these symptoms. The present case is unique because massive ICH resulting from pituitary apoplexy caused severe conscious disturbance.

Rare hemorrhagic patterns caused by pituitary apoplexy are known. SAH associated with pituitary apoplexy is a rare but well-known pattern of pituitary apoplexy. The anatomical proximity of the sellae turcica to the basal cistern may be the cause of this symptom. Extravasation of blood from a bleeding pituitary tumor may enter the subarachnoid space resulting in the clinical presentation of SAH. However, the present case manifested as massive ICH without SAH. Only one previous case report has involved ICH as the result of pituitary apoplexy. In our case, we observed that the tumor was partly embedded in the left frontal lobe. We speculate that necrotic and hemorrhagic changes occurred in this part of tumor leading to ICH. This entity may be difficult to distinguish from ruptured aneurysm of the anterior cerebral artery. However, we could diagnose this rare entity relatively easily in the present case, because we knew that the patient had been treated for non-functioning pituitary adenoma conservatively at another hospital.

Fatal pituitary apoplexy is known in only a few case reports. Hypothalamic compression caused by intratumoral hemorrhage and rapid enlargement of the tumor was considered to lead to unfavorable outcomes. Serious bleeding into the subarachnoid cistern and/or intraventricular space with unfavorable outcomes were also reported. Our patient also suffered a severe clinical course and finally died of massive ICH. Operative indications for asymptomatic non-functioning pituitary adenoma are still controversial, in spite of recent surgical developments for pituitary adenoma including endoscopic surgery. The present case shows that asymptomatic pituitary adenoma may occasionally cause pituitary apoplexy and may result in severe neurological deficit or even death.

References

26) Trumble HC: Pituitary tumours; observations on large tumours which have spread widely beyond the confines of the sella turcica. Br J Surg 39: 7–24, 1951

Address reprint requests to: Kota Kurisu, MD, Department of Neurosurgery, Otaru Municipal Medical Center, 3–11–1 Nagahashi, Otaru, Hokkaido 047–0036, Japan.
e-mail: kota0912kuri@yahoo.co.jp