Case Report

A Case of Apoplexy of Rathke’s Cleft Cyst Followed by Cerebral Infarction

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Rathke’s cleft cyst (RCC) apoplexy is a rare clinical entity. We report a case of apoplexy of an RCC followed by cerebral infarction. A 67-year-old woman was found lying on the street unconscious. She had fallen from her motorbike. On referral to our hospital she gradually regained consciousness and presented with no neurological deficits. CT showed a round and slightly hyperdense area in the suprasellar region. However, the attending physician did not consult a neurosurgeon and the patient was discharged the same day. Thirteen days after the first emergency visit she developed left hemiparesis and dysarthria. CT showed a round hypodense area in the suprasellar region. The change of the density in the suprasellar region on CT suggested the pituitary apoplexy. CT also showed a low density area in the territory of the right middle cerebral artery, which indicated the cerebral infarction. MR angiography revealed poor visibility and stenotic changes of right middle cerebral arteries. Transsphenoidal surgery was performed. Histopathological findings confirmed a hemorrhagic RCC. Postoperative MR angiography showed that the visibility and stenosis of right middle cerebral arteries were recovered. This is the rare case of apoplexy of an RCC followed by cerebral infarction.

1. Introduction

Rathke’s cleft cysts (RCCs) were found frequently (13–22%) in normal pituitary glands at autopsy [1]. Most RCCs are small and asymptomatic throughout life. RCC presents various symptoms when it enlarges to compress the optic chiasm, hypothalamus, and pituitary gland [2–4]. The symptoms include headache, visual disturbance, hypopituitarism, and diabetes insipidus. Apoplexy of an RCC is a very rare clinical entity. Pituitary apoplexy occurs in 0.4–16.6% of all pituitary adenomas [5, 6]. Pituitary apoplexy followed by cerebral infarction is rare. There are 2 possible mechanisms of cerebral infarction in pituitary apoplexy, that is, vasospasm or compression of an artery. This is the rare case of apoplexy of an RCC followed by cerebral infarction.

2. Case Report

A 67-year-old woman was found lying on the street unconscious. She had fallen from her motorbike. On referral to our hospital she gradually regained consciousness and presented with no neurological deficits. She had a facial abrasion, but no headache and no nausea. CT showed a round and slightly hyperdense area in the suprasellar region (Figure 1(a)). However, the attending physician did not consult a neurosurgeon and the patient was discharged the same day. This physician overlooked a significant appearance in the suprasellar region. Thirteen days after the emergency visit, she gradually developed left hemiparesis. Other neurological signs were within normal limits. Arterial blood gas measurement did not reveal acidemia or alkalemia. Routine laboratory tests were normal, and the patient was neither diabetic, nor hypertensive, nor hyperlipidemic. Endocrinological tests also detected no abnormalities. Echocardiographic examination and carotid artery ultrasound examination showed no abnormalities, and the electrocardiogram was also normal.

CT on admission showed a round hypodense area in the suprasellar region (Figure 1(b)). The change of the density in the suprasellar region on CT suggested the pituitary apoplexy. Magnetic resonance imaging (MRI) taken 2 days...
after admission detected isointensity on T1WI and high intensity on T2WI in the suprasellar region (Figure 2). CT also showed a low density area in the territory of the right MCA, which indicated the cerebral infarction (Figure 1(c)). MR angiography (MRA) showed signal loss with poor visibility of distal right middle cerebral arteries (MCAs) (Figure 3). The conservative treatment for cerebral infarction improved her left hemiparesis.

Ten days after admission, endoscopic endonasal trans-sphenoidal surgery was performed. The sella turcica was found to be thin. The tumor contained white-tinged viscid fluid. A normal pituitary gland was observed on the right side. Histopathological findings confirmed hemorrhagic RCC (Figures 4(a) and 4(b)). The cyst wall was lined by a ciliated columnar cell layer with goblet cells. Thin blood vessels were observed in the cyst wall. Red blood cells were seen in both cyst and cyst wall. Postoperative MRI showed the decompression of suprasellar region (Figure 5). Postoperative MRA revealed partial stenotic changes of right MCAs (Figure 6). These findings suggested RCC apoplexy followed by cerebral infarction.

3. Discussion

RCC apoplexy is a rare clinical entity. Fourteen cases of apoplexy of RCC have been reported in the literature (Table 1) [6–17]. The mechanism of RCC apoplexy is supposed to be the repeated minor bleeding from the thin blood vessels in the cyst wall by the stimulation of cyst contents and the bleeding from the hypophyseal portal blood vessels by the compression or shearing stress [7, 15]. The clinical presentations of these cases were headache, nausea, visual disturbance, and cranial nerve palsy. One case in Table 1 presented with altered consciousness as our case did. Nawar et al. reported 11 cases with hemorrhage within RCC [14]. In their study, although not described in detail of each case, respectively, there were 3 patients with altered consciousness, there were 10 patients with headache, there were 3 patients with a visual deficit, and there was 1 patient with cranial nerve palsy.

The pituitary apoplexy is caused by the anticoagulant therapy, the bromocriptine therapy, the radiation therapy, the hormone loading test, the cerebral angiography, and the cardiovascular surgery [18]. Particularly in macroadenoma, the head trauma can cause the apoplexy between 0 days and 21 days [19–21]. None of these reviewed cases of RCC apoplexy were caused by the head trauma. In our case, it was unclear whether the unconsciousness at the first emergency visit depended on the head trauma or apoplexy.

Preoperative endocrinological evaluations revealed abnormalities in 4 cases (Table 1). Wakai et al. described that preoperative endocrinological examinations revealed abnormalities in 3 cases. Most pituitary apoplexy cases need hormone replacement therapy for partial hypopituitarism [6]. In our case the preoperative endocrinological examinations were normal.

A rare complication of pituitary apoplexy is cerebral infarction, which is caused by either direct compression of an artery or vasospasm. Twenty cases of pituitary apoplexy followed by cerebral infarction have been reported in the literature (Table 2) [1, 20, 22–24, 26, 28, 29, 31–39]. The ischemic events were attributed to mechanical compression by the tumor in 12 cases and to cerebral vasospasm in 8 cases. The cerebral infarction was located at the anterior cerebral artery (ACA) territory in 4 cases, the MCA territory in 8 cases, and the ICA territory in 4 cases. Cerebral arterial stenosis was detected in the ICA in 15 cases, in the MCA in 3 cases, and in the ACA in 3 cases. The cerebral infarction occurred between 0 and 21 days after the onset of pituitary apoplexy. Most infarctions due to cerebral vasospasm occurred between 5 and 21 days after onset, and all infarctions by mechanical compression occurred within 2 days after onset.

In our case, the cerebral infarction occurred in the right MCA territory after a 13-day interval from the onset of RCC apoplexy.
Figure 2: Axial, sagittal, and coronal views of the MRI showed isointensity on T1WI (a, e, and g) and high intensity on T2WI (b, f, and h) in the suprasellar region. Axial and coronal views of the MRI presented islow intensity on T1WI (c, g) and high intensity on T2WI (d, h) in the territory of the right middle cerebral artery.
Figure 3: MRA revealed less visibility and stenotic changes of right middle cerebral arteries.

Figure 4: The cyst wall shows ciliated columnar epithelium with goblet cells. Thin blood vessels were observed in the cyst wall. Red blood cells were seen in both cyst and cyst wall (H&E original magnification: (a) ×200; (b) ×400).

Table 1: Summary of the clinical presentations and endocrinological findings in reported cases of RCC apoplexy.

| Author and year | Age, sex | Presentation | Preop endocrine findings | Postop endocrine results |
|-----------------|----------|--------------|--------------------------|--------------------------|
| Onesti et al., 1990 [16] | 25, F | Headache, nausea | Normal | Normal |
| Kleinschmidt-DeMasters et al., 1995 [11] | 51, F | Visual deterioration | NA | NA |
| Kurisaka et al., 1998 [18] | 8, F | Headache | Normal | Normal |
| Nishioka et al., 1999 [15] | 46, F | Headache, visual loss, nausea | Normal | Normal |
| Fukushima et al., 2001 [22] | 67, F | Headache, nausea, ptosis | Normal | HRT for cortisol |
| Pawar et al., 2002 [17] | 19, M | Headache, blurred vision | Normal | Normal |
| Rosales et al., 2004 [23] | 34, M | Headache, diplopia | PRL elevation, decreased T4 | HRT for DI and thyroid |
| 24, F | Headache | Normal | Normal |
| 20, M | Headache, nausea, diplopia | Decreased testosterone | HRT for testosterone |
| Binning et al., 2008 [9] | 23, F | Headache, visual loss | PRL elevation | Normal |
| 49, M | Headache | Normal | Normal |
| 21, F | Headache | Decreased T4 | HRT for thyroid |
| 54, F | Headache, visual loss | Normal | Normal |
| Raper and Besser, 2009 [24] | 72, F | NA | NA | NA |
| Present case | 67, F | Hemiparesis | Normal | Normal |

PRL, prolactin; DI, diabetes insipidus; HRT, hormone replacement therapy; NA, not available.
Figure 5: Sagittal and coronal views of the MRI showed the decompression of suprasellar region. (a, c) T1WI and (b, d) T2WI.

Figure 6: Postoperative MRA revealed partial stenotic changes of right MCAs.

apoplexy. MRA revealed the stenotic lesion of the right MCA, and echocardiogram, electrocardiogram, and carotid artery ultrasound showed no abnormalities. These clinical findings could exclude the embolic cause of the ischemic lesion. Our case was neither hyperlipidemic nor hypertensive. CTA and carotid artery ultrasound presented no artherosclerotic changes. These clinical findings could exclude the possibility of the artherosclerotic brain infarction. Postoperative MRA and CTA showed that the visibility and stenosis of right MCAs were recovered. Therefore together these findings suggested that the etiology of the cerebral infarction was cerebral vasospasm.
Table 2: Reported cases of cerebral ischemia following pituitary apoplexy.

| Author and year                | Age, sex | Territory of infarction | Angiographical findings | Symptom                        | Days after onset | Mechanism | Pathology |
|--------------------------------|----------|-------------------------|-------------------------|-------------------------------|-----------------|-----------|-----------|
| Rosenbaum et al., 1977 [25]    | 77, M    | Right MCA               | Right ICA occlusion, left ICA stenosis | Left hemiparesis             | 0               | c         | PA        |
| Cardoso and Peterson, 1983 [19]| 34, F    | Diffuse                 | Bil. ICA, ACA, MCA stenosis | Reduced consciousness        | 21              | v         | PA        |
|                               | 38, M    | NA                      | Bil. ICA, BA stenosis    | Lethargic                    | 0               | v         | PA        |
| Bernstein et al., 1984 [8]     | 48, M    | NA                      | Bil. ICA stenosis        | Reduced consciousness, hemiparesis, left hemiparesis | 0               | c         | PA        |
| Clark et al., 1987 [26]        | 40, M    | Left ACA                | Right ICA stenosis, left ICA occlusion | Dysphasia, right hemianopia, right hemiparesis | 0               | v         | PT        |
| Pozzati et al., 1987 [21]      | 15, M    | Right MCA               | Bil. ICA occlusion       | Reduced consciousness        | 0               | v         | PT        |
| Itoyoama et al., 1990 [27]     | 45, M    | NA                      | Left ICA, MCA stenosis   | Reduced consciousness, right hemiparesis | 14              | v         | PA        |
| Yaghmai et al., 1996 [28]      | 47, M    | None                    | Right ICA occlusion      | Right blindness              | 0               | c         | PA        |
| Lath and Rajeshkhar, 2001 [12] | 40, M    | Right ICA               | Right ICA occlusion      | Left hemiparesis             | 1               | c         | PA        |
| Rodier et al., 2003 [29]       | 35, M    | Right ICA, ACA          | Bil. ACA, right MCA stenosis | Reduced consciousness, left hemiparesis, reduced consciousness | 2               | c and v   | PA        |
| Akutsu et al., 2004 [30]       | 29, M    | Left MCA                | Left ACA stenosis        | Reduced consciousness, right hemiparesis | 5               | v         | PA        |
| Byung et al., 2007 [10]        | 41, M    | Left MCA                | NA                      | Right hemiparesis, dysarthria | 7               | v         | PA        |
| Dogan et al., 2008 [31]        | 50, M    | Left ICA                | Left ICA occlusion       | Reduced consciousness        | 0               | c         | PA        |
| Das et al., 2008 [32]          | 46, M    | Left MCA                | Left ICA stenosis        | Right hemiparesis             | 0               | c         | PA        |
| Ahmed and Semple, 2008 [33]    | 51, M    | Bil. ACA                | NA                      | Left hemiparesis             | 0               | c         | PA        |
| Yang et al., 2008 [34]         | 43, M    | Right ICA               | Right ICA stenosis       | Left hemiparesis             | 0               | c         | PA        |
| López Hernández, 2008 [35]     | 23, M    | —                       | —                       | —                             | —               | —         | PT        |
| Lill et al., 2009 [13]         | 59, M    | Bil. ICA                | Bil. ICA occlusion       | Left hemiparesis, reduced consciousness | 0               | c         | PA        |
| Chokyu et al., 2011 [36]       | 50, M    | Right MCA               | Right ICA occlusion      | Left hemiparesis             | 0               | c         | PA        |
| Present case                   | 67, F    | Right MCA               | Right MCA stenosis       | Reduced consciousness, left hemiparesis | 13              | v         | RCC       |

ACA, anterior cerebral artery; MCA, middle cerebral artery; ICA, internal carotid artery; c, compression; v, vasospasm; PA, pituitary adenoma; PT, pituitary tumor.

The pathology of vasospasm following pituitary apoplexy is unclear; however, vasoactive chemical substances released during pituitary apoplexy may cause vasospasm [20, 28, 35]. The subarachnoid hemorrhage (SAH) causes the vasospasm, but the pathological mechanism also remains unclear [27]. The endothelin, angiotensin, and sphingosine metabolites are one of the vasoactive substances, respectively [5, 25, 30]. Although the CT and MRI detected no SAH in our case, vasoactive substances liberated from the hemorrhagic RCC or SAH below levels in detection by CT and MRI might cause the vasospasm.

This patient had fallen from her motorbike. She was disoriented and had a facial abrasion. Considering head trauma, the attending physician examined head CT. This physician...
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