Pituitary apoplexy as a consequence of lymphocytic adenohypophysitis in a pregnant woman: a case report

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Objective and importance: A patient with pituitary apoplexy resulting from lymphocytic adenohypophysitis, which caused visual disturbance during pregnancy, is described. This is the first report of such case.

Clinical presentation: A 23-year-old primigravida in her 25th week of gestation experienced headache and bitemporal hemianopsia of sudden onset. Magnetic resonance imaging (MRI) revealed a large pituitary mass with intratumoral hemorrhage. Although conservative treatment with intravenous glycerol improved the symptoms partially, the visual symptoms worsened again 6 weeks later. After delivering a girl by scheduled caesarean section her visual symptoms improved. Despite the symptomatic improvement, MRI showed the chiasmatic compression by the enlarged pituitary gland had not changed. Therefore, trans-sphenoidal surgery to decompress the chiasm was performed. Necrotic tissue was seen exuding behind the enlarged pituitary gland and adenohypophysitis with bleeding (apoplexy) was diagnosed histologically. After follow-up for 40 months, she was doing well without any visual or neurological deficits.

Conclusion: Although relatively rare, pituitary apoplexy as a consequence of lymphocytic adenohypophysitis should be borne in mind when a pregnant woman presents with headache and visual disturbance of sudden onset. [Neurol Res 2005; 27: 399–402]

Keywords: Pituitary apoplexy; lymphocytic adenohypophysitis; pregnancy

INTRODUCTION
Lymphocytic adenohypophysitis is a rare event and symptoms due to pituitary gland enlargement, including headache, nausea and vomiting, as well as visual field deficits have been documented. However, to our knowledge, there have been no previous descriptions of pituitary apoplexy as a consequence of lymphocytic adenohypophysitis occurring during pregnancy and causing acute-onset chiasmatic compression. Here, we describe such a patient, who developed visual disturbance during her pregnancy. Neuroradiological and histological examinations confirmed the diagnosis of bleeding in the area of lymphocytic adenohypophysitis. This pathological condition should be borne in mind when a pregnant woman presents with acute-onset visual symptoms.

CASE REPORT
A 23-year-old primigravida in her 25th week of gestation had a 1-week history of retro-orbital pain and headache, which suddenly worsened 4 days prior to visiting an ophthalmologist at a local hospital. Bitemporal hemianopsia was found and a magnetic resonance imaging (MRI) scan showed a large mass occupying the pituitary fossa and suprasellar cistern. She was transferred and admitted to Teikyo University Hospital immediately for further treatment on 27 July 2000.

On admission, her visual acuity was 0.5 for the right eye and 0.1 for the left. The central scotoma was enlarged and temporal hemianopsia of the right eye was documented. None of the pituitary hormones was abnormally elevated with the exception of prolactin, which was slightly elevated (36.0 ng/ml). She did not have diabetes insipidus. MRI scan showed a mass lesion in the pituitary fossa that extended into the suprasellar cistern and compressed the optic chiasm. T1-weighted image showed an iso-intense to slightly low-intensity lesion and T2-weighted images showed a lesion with markedly high signal intensity in the dorso-superior part of the enlarged pituitary gland (Figure 1A–C). Gadolinium enhancement was not done because of government regulation of contrast media for pregnant women. Therefore, the lesion was diagnosed as a pituitary adenoma with apoplexy. Since the patient
was pregnant, conservative treatment with 200 ml intravenous glycerol twice daily and an oral hydrocortisone supplement was started. Her visual symptoms did not worsen further and her headache subsided. Follow-up MRI scans on 4 August showed that the signal intensity of the T1-weighted image of the lesion had increased and that part of the T2-weighted image was low, thus confirming the presence of hemorrhage. She was discharged on 8 August 2000 on an oral hydrocortisone supplement (10 mg/day).

On 14 September 2000 (week 32 of gestation), her bitemporal hemianopsia worsened and she was re-admitted. An elective caesarean section was planned for the 34th gestational week and she delivered a baby girl on 29 September 2000. Although her visual acuity improved and her visual field widened after delivery, MRI showed that the optic chiasm was severely compressed by the mass (D) and the signal intensity of the hemorrhagic lesion had become high on the T1-weighted image (E), and a mixture of low and high on the T2-weighted image (F).

Figure 1: MRI scan on 21 July, 2000 (A,B,C). The pituitary gland was swollen and formed a mass lesion extending from the pituitary fossa to the suprasellar area. The T1-weighted image showed an iso-intense to slightly low-intensity lesion and T2-weighted images showed a lesion with markedly high signal intensity in the dorso-superior part of the enlarged pituitary gland (A,B,C), suggesting recent hemorrhage. Follow-up MRI on 4 August, 2000 (D,E,F). On T1-weighted coronal images, the optic chiasm was severely compressed by the mass (D) and the signal intensity of the hemorrhagic lesion had become high on the T1-weighted image (E), and a mixture of low and high on the T2-weighted image (F).

HISTOLOGICAL EXAMINATION
The tissue of the biopsy specimen was infiltrated by lymphocytes. Hemosiderin deposition between the necrotic tissue and pituitary gland was observed, and a surgical probe, glue-like, liquefied, necrotic tissue was seen exuding from behind it. A small biopsy specimen of the surrounding pituitary tissue was taken, frozen and examined intra-operatively, and this revealed lymphocytic infiltration. Lymphocytic adenohypophysitis was considered and the surgery was finished after curettage of the necrotic tissue. The patient’s post-operative course was uneventful, and she was discharged without any visual or neurological symptoms. Endocrinological stimulation tests with corticotrophin-, thyrotrophin-, growth hormone- and luteinizing hormone-releasing hormones were performed on 25 October and showed a slightly impaired corticotropin response, but the other pituitary hormones responded well to stimulation. An immunological examination performed afterwards did not show any auto-immune responses, such as elevated rheumatoid factor or antinuclear antibody titers. Three years after surgery, she was well with mild hypopituitarism, which was well controlled with minimal oral hydrocortisone replacement therapy (10 mg daily).
confirming that the pituitary apoplexy had occurred as a consequence of lymphocytic adenohypophysitis during pregnancy (Figure 2).

**DISCUSSION**

Lymphocytic adenohypophysitis is a rare event, but most such events happen during pregnancy or the postpartum period. Symptoms due to pituitary gland enlargement, including headache, nausea and vomiting, as well as visual field deficits, have been documented in approximately 40–60% of patients. However, to our knowledge, no descriptions of pituitary apoplexy as a consequence of lymphocytic adenohypophysitis occurring during pregnancy or causing acute-onset chiasmatic compression have been published. Pituitary apoplexy occurring together with adenohypophysitis is also a very rare event and only one such case, a patient with adenohypophysitis associated with polyglandular syndrome, has been reported.

The etiology of pituitary apoplexy due to pituitary adenoma is not fully understood. Various predisposing factors, such as anticoagulant use, head trauma, bromocriptine treatment and endocrine stimulation tests have been reported, and pregnancy itself has also been reported to be a predisposing factor. It would appear that a poor blood supply to the tumor or fragile blood vessels within it lead to ischemia and hemorrhagic infarction or bleeding.

In our patient, the enlarged pituitary gland infiltrated by lymphocytes might have needed an increased blood supply and thus mimicked a pituitary adenoma. As described above, pregnancy is also another condition that predisposes to pituitary apoplexy. Therefore, these two conditions together might have led to pituitary apoplexy in this patient.

Initially, our patient’s symptoms during pregnancy that were caused by pituitary apoplexy were relieved by conservative therapy with intravenous glycerol. The oral corticosteroid given to maintain a normal glucocorticoid level might also have helped relieve the symptoms, as resolution of adenohypophysitis by glucocorticoid therapy has been reported. However, the visual symptoms that initially improved with conservative therapy worsened as her pregnancy progressed. Previous reports have suggested that pituitary swelling due to adenohypophysitis might not change during pregnancy and might subside spontaneously in some cases that are followed up for longer. Our patient’s visual disturbance improved after delivery, but the neuroradiological findings of severe chiasmatic compression and the preoperative diagnosis of pituitary apoplexy, we decided to perform surgery. After curettage of the necrotic tissue, her symptoms resolved completely. Hypopituitarism associated with lymphocytic adenohypophysitis, especially in patients with a longer history, has been documented. Our patient,

![Figure 2: Light microscopic appearance of the histological specimen obtained during trans-sphenoidal surgery (hematoxylin and eosin staining, objective lens at ×10 (A) and ×20 (B–D)]. At both the high and low magnifications, marked lymphocytic infiltration of the pituitary gland was evident (A,B). A necrotic area and nearly necrotic cells adjacent to it were observed (C). Hemosiderin deposition was observed in the necrotic area, confirming pre-existing hemorrhage (D).
however, had only mild hypopituitarism 3 years after surgery. This might be because we removed only the necrotic tissue and left the normal gland, as histological examination of frozen sections during surgery excluded the presence of a pituitary adenoma.

**CONCLUSION**

A patient with pituitary apoplexy resulting from lymphocytic adenohypophysitis, which caused visual disturbance during pregnancy, is described. Although relatively rare, pituitary apoplexy resulting from lymphocytic adenohypophysitis should be borne in mind when a pregnant woman presents with headache and visual disturbance of sudden onset.

**REFERENCES**