Case reports

Spontaneous regression of pituitary mass in temporal association with pregnancy

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Summary. We have encountered a case of a pituitary mass which emerged and enlarged during pregnancy in a 29-year-old woman. On CT scanning the mass disappeared over the course of four months postpartum and was followed by pituitary hypofunction. The hypofunction was restricted to ACTH, GH, and PRL. The visual field defects, bitemporal hemianopsia, disappeared with disappearance of the pituitary mass on CT scanning, indicating that such deficits during this period were reversible. Based on the clinical course and laboratory data, this case is thought to have been lymphocytic adenohypophysis. It was concluded that among pituitary tumors developing during pregnancy there are some such cases which do not require surgical therapy.

Key words: Pituitary tumor - Pregnancy - Lymphocytic adenohypophysis - Hypopituitarism

Case report

The patient was a 29-year-old woman of medium physique complaining of headache and disturbances of the visual field. Her medical history included myocarditis at the age of 10 when she was hospitalized for one month. She was married at the age of 25 and had had no previous pregnancies or experience of lactation.

The last menstruation was August 30, 1983, and she was found to be pregnant on September 10. With progressing pregnancy, there was enlargement of the mammary glands and some lactation, but by the eighth month of pregnancy, the mammary glands had begun to shrink and there was no galactorrhea.

At the start of the ninth month of pregnancy, she experienced a left frontal headache, together with nausea and vomiting, and during the ninth month she noted difficulties in the right visual field. Thereafter, the headache and visual field defects worsened, but by the end of the ninth month, the headache and nausea had disappeared. On May 15, 1984, there was normal delivery of a 3100 g boy, but, on the third day postpartum, the frontal headache returned more severely. Complaining of visual field defects, she was admitted to our clinic on May 25.

On admission, consciousness was normal. Blood pressure was 102/62 mmHg and heart rate 72/min. Visual acuity was reduced to 50 cm m/m. (0.03 x -2.0 D) on the right and 0.5 (1.0 x -1.0 D) on the left, and examination of the visual fields revealed bitemporal hemianopsia (Fig. 1a). No other neurological deficits were found, but lactation was not evident despite the fact that it had been only 10 days from parturition.
Serum electrolytes, hepatic function and renal function were normal. In periferal blood, there was a leucocytosis (11700/mm³).

Although thinning of the posterior clinoid process was seen in plain skull X-rays, there was neither a double floor nor sellar enlargement. CT scans showed a mass extending superiorly from the sella turcica with homogenous contrast enhancement (Fig. 2a, b). In cerebral angiograms, there was superior displacement of both A1 portions and a primitive trigeminal artery leading from the right internal carotid artery to the basilar artery (Fig. 3a, b).

Due to vertigo, general fatigue and decreased blood pressure one month after delivery, the patient was started on adrenocorticoioid hormone supplement therapy (Predonin, 10 mg/day). While awaiting surgery under a diagnosis of pituitary tumor, CT scans showed reduction in the pituitary mass two months after delivery and improvements in the visual field deficits. While observing the course of the disease, visual acuity improved to 0.6 (1.2 x -1.0D) on the right and 0.8 (1.2 x -1.0D) on the left and the visual field became normal three months from parturition (Fig. 1b). The pituitary mass was no longer visible in CT scans (Fig. 4). Hormone supplement therapy was halted and a 7-day menstruation resumed on August 8, 1984. Thereafter, menstruation occurred once every four months, but no lactation was evident for the entire postpartum period.

During this time, she experienced muscular pain in the arms and was unable to raise them to shoulder height. Six months from delivery, hormone supplement therapy (Hydrocortisone, 30 mg/day) was resumed due to loss of appetite, general fatigue, nausea and a marked loss of body weight. Endocrinological tests of pituitary function revealed low or absent responses to hormone stimulation (ACTH, GH and PRL) (Table 1).

On December 18, 1985 she showed symptoms of a common cold and a fever of 40°C. On the following day, she suddenly convulsed for 30 s with the arms flexed and legs extended. Since such convulsions occurred three times and there were no notable changes in CT scans, she was given anticon-
Fig. 2a and b. Enhanced CT scans 26 days after delivery showing a homogeneously contrast-enhanced mass extending into suprasellar cistern. a axial view, b coronal view

tulous therapy. She was admitted to hospital on June 20, 1986 for further examination.

On re-admission, consciousness was clear, body temperature was 36.5°C, blood pressure was 90/52 mmHg and general fatigue was apparent. ESR level was increased to 25/70, WBCs were 5200/mm³ and CRP was negative. Urinary sugar and protein were absent. Basal levels of pituitary hormones are shown in Table 1, and there were no circadian rhythm in ACTH-cortisol. She was positive for anti-nuclear antibodies, which were of the speckled type, but anti-RNP antibody, anti-Sm antibody and SS-A/SS-B antibody were negative.

Lymphocyte subsets were normal, with OKT4 = 46.2% and OKT8 = 24.2%. Immunoglobulin fractions were normal. HLA typing was A2,B6, CW1,CW3. Elevation of the MB fraction was seen in CPK isozyme. Pulmonary function tests showed a decrease in %VC to 69.9% and a disturbed pattern of respiratory contraction. EEG showed diffuse slow waves with dominant delta waves.

Discussion

Lesions presenting as the chiasmal syndrome, in which a pituitary mass is detected in association with pregnancy or delivery, suggest one of three possible conditions: (1) physiological enlargement of the pituitary gland during pregnancy, (2) prolactinoma, or (3) lymphocytic adenohypophysitis.

It is well-known that the mass of the pituitary gland can increase during pregnancy and Erdheim [2] maintains that it may increase as much as 70%. The cause for such physiological enlargement is primarily an increase in prolactin-secreting cells of the anterior pituitary due to the massive increase in estrogen from the feto-placental unit [3]. It is said that, following delivery, the headache and visual field deficits ameliorate [4].

In human prolactinoma, there is also a high concentration of estrogen receptors [5] and in a state with high levels of estrogen, such as in pregnancy, it is possible that the prolactinoma would develop further [6-8]. Since the blood estrogen until delivery continues at a high level, it is thought that there is proliferation of the prolactin cells or prolactinoma and enlargement proportional to the weeks of pregnancy. At delivery, there is a sharp decrease in blood estrogen and a concomitant fall in blood prolactin. In prolactinoma cases, there is also a fall in blood prolactin following delivery and it is thought that estrogen stimulation does not bring about persistent growth of the adenoma [9]. For this reason, the chiasmal syndrome seen in prolactinoma cases and in physiological enlargement of the pituitary is thought to ameliorate following delivery [10].

In lymphocytic adenohypophysitis, it is said that the symptoms worsen following delivery [11]. In our case, immediately prior to delivery, there were transient improvements in the symptoms of the chiasmal syndrome which had appeared during the ninth month of pregnancy. Following delivery, however, there was worsening of symptoms and a clinical course which differs distinctly from that found in
prolactinoma or the physiological enlargement of the pituitary.

It is well-known that, together with increase in blood prolactin levels, galactorrhea occurs, but in the present case there was enlargement of the mammary glands only until the eighth month of pregnancy and then shrinkage quite different from galactorrhea. Normally, prolactin levels decrease gradually following delivery and after 2–3 months are at a level similar to the pregnancy state, but in our case the prolactin levels in blood obtained on the 16th day postpartum was less than 2.0 μg/ml, which is a low value given the proximity to delivery. It is difficult to accept that these findings indicated that blood prolactin levels were high during the enlargement of the tumor.

In research on the pituitary hormone-secreting capacities in prolactinoma cases, GH is found to be most easily influenced [12, 13]. In macroadenoma, 90–100% of GH secretion is disturbed, whereas ACTH is least easily influenced. In our examination of the pituitary secreting capacities of the present case, low responses of ACTH, GH and PRL were seen – a pattern which differs from that seen in prolactinoma cases.

The first case of lymphocytic adenohypophysitis was reported by Goudie in 1962, as a disorder associated with Hashimoto's disease in an autopsy case.
The clinical course of such cases has been described as follows. The patients present with the chiasmal syndrome during the latter part of pregnancy and there is sudden disappearance of the pituitary mass following delivery. Within one year postpartum there is a decrease in pituitary function in most cases [1, 14–17]. The emergence of the chiasmal syndrome is said not always to parallel the course of the pregnancy and delivery. Near the end of pregnancy there may be temporary improvements of symptoms and aggravation immediately following delivery. The pituitary hypofunction usually includes disturbances of ACTH, whereas LH and FSH are relatively preserved [14–18].

From the above findings it is evident that the clinical course in our patient was similar to that reported to be typical of lymphocytic adenohypophysitis. Although histopathological study has not yet been performed, Zellar [10] and Aida et al. [19] have reported cases with clinical courses similar to lymphocytic adenohypophysitis and we consider our patient also to have been one such rare case in which the natural history of a pituitary tumor could be followed.

While it is known that, following the disappearance of the pituitary tumor in such cases, pituitary hypofunction remains, we believe it is appropriate not to pursue surgical treatment immediately, but to give hormone supplements while closely observing the course of the disease both clinically and on serial CT scan.

In conclusion, when findings which are atypical of prolactinoma and suggestive of autoimmune disease are found in cases with a pituitary mass during pregnancy or following delivery, it is rational to observe the clinical course carefully and carry out serial CT scanning.

References


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