Successful pregnancy in a patient with pre-existing lymphocytic hypophysitis

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Lymphocytic hypophysitis (LyH) is a rare disorder characterized by lymphocytic infiltration of the pituitary gland invariably resulting in pituitary hormone deficiency. LyH is usually diagnosed at surgery in young women during late pregnancy or at the post partum period. The effect of a subsequent pregnancy on the course of the disease is not known. We describe the first case of pregnancy and delivery in a woman with established LyH and partial hypopituitarism.

Case report

A 21 year old woman (gravida 3, para 3) developed severe frontal headaches with a progressive decrease in the peripheral vision of her left eye during her third pregnancy and worsening of the symptoms seven months post delivery. She initially breast fed, but lactation ceased abruptly after four months and menses did not recur. Her past medical history was unremarkable except for headaches during her previous two pregnancies that resolved soon after delivery. Physical examination revealed an apparently healthy woman with normal vital signs. Ophthalmologic examination showed bilateral decreased visual acuity with asymmetrical loss in both temporal aspects of her visual fields. A brain CT scan revealed a 2.5 cm pituitary mass with suprasellar extension and a central area of lower density compatible with hemorrhage or necrosis. The serum biochemistry and complete blood count were normal; the sedimentation rate was 50 mm/hour. Basal plasma prolactin, thyroid hormone, cortisol and gonadotropin levels were within normal limits. Due to the visual impairment, the patient underwent transphenoidal surgery – no adenoma was found and a partial hypophysectomy was performed. Histological examination revealed epithelial cells surrounded by a dense infiltrate of lymphocytes and plasma cells, few eosinophils and widespread fibrosis, consistent with lymphocytic hypophysitis. Following surgery, visual acuity and visual fields improved, as did the headache. In an attempt to suppress the pituitary inflammatory process, she was treated with prednisone 60 mg daily with gradual tapering to 5 mg daily over the next six months. A Repeat CT scan 5 months after surgery revealed a 1.5 cm sellar mass. Three months after surgery, menses resumed and five

Abbreviations:
LyH: lymphocytic hypophysitis; TSH: thyroid stimulating hormone; ACTH: adrenocorticotropic; TRH: thyrotropin releasing hormone; GH: growth hormone; LH: luteinizing hormone; FSH: follicle stimulating hormone; MRI: magnetic resonance imaging; CT: computerized tomography; VEP: visual evoked potential.
months later she conceived spontaneously and discontinued the treatment with prednisone. She was referred to our hospital in the second month of pregnancy in good general condition except for recurrent nausea and vomiting. The physical examination was again unremarkable and evaluation of pituitary function revealed hypopituitarism with deficiencies of thyroid stimulating hormone (TSH), prolactin and cortisol (Table I). The patient was treated with 5 mg of prednisone and 100 μg of L-thyroxine daily and was followed periodically with visual acuity, visual fields and visual evoked potential (VEP) evaluation. At 32 weeks of pregnancy, due to severe headache and suspicion of apoplexy, MRI of the pituitary was performed which, surprisingly, showed no residual mass with a small, atrophic appearing pituitary. At 39 weeks of gestation a healthy 2,840 gr. male infant was delivered by uncomplicated vaginal delivery. The patient failed to lactate post partum. Repeated pituitary function tests three months after delivery showed no improvement of thyrotroph, corticotroph and lactotroph function with sparing of the gonadotroph ovary axis (Table I).

Discussion
Lymphocytic hypophysitis is a rare disorder characterized by lymphocytic infiltration of the pituitary gland that occurs primarily in women during pregnancy or the post partum period (1–5). The clinical manifestations of LyH are due to the pituitary lesion mass effect, namely, headache and visual field defects or due to pituitary hormone deficiency. During the last decade, LyH has been increasingly recognized as a cause of hypopituitarism and its actual incidence is probably higher than currently recognized, since asymptomatic patients with small sellar masses and those with idiopathic pituitary hormonal deficiencies are not diagnosed.

The etiology of LyH is unknown, but it is thought to be an autoimmune disorder. This is supported by its propensity to occur in women, the presence of anti-pituitary antibodies in the sera of some patients, the association with other autoimmune disorders (thyroiditis in particular) and the selective, at times reversible, pituitary hormonal deficiencies that cannot be explained by simple destruction of the gland (2). Although all pituitary hormones can be affected, the most common endocrine abnormalities are low ACTH and TSH secretion with sparing of the gonadotroph function, as demonstrated by our patient (1). The prolactin levels can be either high or low. In contrast, in patients with large pituitary tumors, gonadotropins and GH secretion are the first to be affected, followed by ACTH and TSH deficiency with prolactin deficiency being rare. When LyH is clinically suspected, a trial of steroids has been suggested to avoid unnecessary surgery (4, 5). As in most patients with LyH, our patient was diagnosed by biopsy and was subsequently treated for 6 months with prednisone. The restoration of her menses and regression of the pituitary mass were most probably due to glucocorticoid therapy, although spontaneous remission cannot be excluded. There is a close association between pregnancy and the initial presentation of LyH, but there are no data on the natural history of the disease in a subsequent pregnancy. Thus, the possibility of aggravation of LyH during subsequent pregnancy is of concern to both patients and clinicians. Our patient conceived spontaneously and her established LyH did not show any apparent adverse influence on her pregnancy, nor did the pregnancy have an adverse effect on the course of her disease. Moreover, resolution of the pituitary lesion was noted. Therefore, LyH should not be considered a contraindication to pregnancy, providing that the judicial use of corticosteroids is contemplated with close neuro-ophthalmological and endocrine follow-up.

References
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CASE REPORT

Abnormal fetal behavior and cardiotocography associated with a congenital meningo-encephalitis

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There are several causes of atypical fetal heart rate patterns. We report a case with highly abnormal cardiotocography associated with a congenital viral meningoencephalitis which was initially mistaken for fetal brain death.

Clinical report

An 18-year-old woman presented to the delivery suite at 38 weeks’ in her first pregnancy with a 48 hour history of absent fetal movement. She also reported a seven day history of malaise, diarrhea and a maculopapular rash. She was not taking any prescribed or recreational drugs. On examination she was apyrexial and her blood pressure was normal. The fundal height was appropriate for gestation and the uterus was not tender. Cardiotocography for 60 minutes revealed a baseline tachycardia of 165 b.p.m. with low variability and no accelerations or decelerations (Fig. 1). At scan the fetus was normally grown and there was no evidence of hydrops or structural anomaly. There was a normal volume of liquor but no fetal limb or breathing movements were seen during a 45 minute period of observation. Maternal investigations revealed a normal hemoglobin (10.3 g/dl), white cell count (11.3×10^6/dl), platelet count (367×10^6/dl), C-reactive protein serum concentration (<6 mg/l), and serum liver enzyme concentrations.

After careful discussion, it was decided that in the absence of a clear diagnosis the fetus should...