Endocr (2011) 39:296–297
DOI 10.1007/s12020-011-9458-1

LETTER TO THE EDITOR

Spontaneous pregnancy in a patient with a relapse of lymphocytic hypophysitis successfully treated with azathioprine and glucocorticoids

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Published online: 27 March 2011

To the Editor,

We report a rare case of recurrent lymphocytic hypophysitis (LyH) after transsphenoidal surgery and high dose methylprednisolone pulse therapy (HDMPT). The patient was treated with azathioprine, and achieved spontaneous pregnancy.

A 22-year-old nulliparous female was seen in June 2004 due to headaches, diplopia and left eye ptosis, and bilateral milky nipple discharge. Six months prior she had undergone transsphenoidal pituitary tumor resection at another hospital, and the histopathological diagnosis was "pituitary tumor with inflammatory response." The patient had no significant medical history, menarche at age 16, and regular cycles until March 2004.

Physical examination was normal except for left eye ptosis and adduction limitation, and bilateral milky nipple discharge. Routine laboratory tests and T3, T4, TSH, LH, FSH, E2, prolactin, ACTH, cortisol, and 24 h urinary free cortisol were within normal limits. Thyroid autoantibodies (TGAb and TPOAb) and TSH receptor antibody were negative. Magnetic resonance imaging (MRI) showed abnormal, enhancing, irregular soft tissue signals with unclear boundaries in the sellar region and suprasellar cistern. Histopathological examination revealed lymphocyte, histiocyte, and neutrophil infiltration in the pituitary tissue, consistent with LyH.

High dose methylprednisolone pulse therapy (HDMPT) was begun, and her symptoms rapidly improved. MRI showed the pituitary significantly reduced in size and the lesions had disappeared. The patient was asymptomatic until December 2005 when she experienced headaches, diplopia, and nausea and vomiting. MRI showed normal hypothalamic and pituitary morphology, and abnormal enhancement on the right side of the dorsum sellae and near the tentorium cerebelli. She was treated with azathioprine 100 mg/day for 16 weeks, and prednisone 30 mg/day (decreased 5 mg/week until withdrawal). After treatment, she experienced regular menstrual cycles and conceived spontaneously in April 2009. Her pregnancy and delivery were uneventful with no LyH relapse during pregnancy or to date.

The natural course of LyH can include sellar mass-occupying effects, pituitary disorders, diabetes insipidus, and hyperprolactinemia [1]. MRI findings are not specific enough to distinguish LyH from pituitary adenomas; thus careful correlation with clinical and laboratory findings is critical [1].

Transsphenoidal resection of pituitary lesions, drugs (corticosteroids, azathioprine, and methotrexate), and radiotherapy have all been used to treat LyH; however, none are effective in all cases [1, 2]. Lecube et al. [3] first reported the successful treatment of LyH using azathioprine in a patient who failed corticosteroid treatment, and subsequently other authors have reported success with azathioprine [4].

Lymphocytic hypophysitis (LyH) is significantly correlated with pregnancy; of 210 reported cases in females, 120 (57%) occurred during pregnancy or the postpartum period, primarily in the final month of pregnancy or first 2 months postpartum [5]. If onset is during pregnancy, complications or adverse effects on the fetus are unlikely, and the condition may gradually resolve after delivery. A history of pregnancy-related LyH is not associated with relapse

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during subsequent pregnancies, and pregnancy can occur in patients with a history of LyH [5].

In summary, LyH is often misdiagnosed as a pituitary tumor. This case suggests that azathioprine and glucocorticoids may reduce the chance of recurrence and increase the chance of spontaneous pregnancy.

Conflict of interest The authors declare that they have no conflict of interest.

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