中文題目：發生於一對姊妹的產後腦下腺功能不足症

英文題目：Postpartum hypopituitarism in two siblings

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Background: Postpartum hypopituitarism, also called Sheehan's syndrome, is caused by intrapartum or postpartum hemorrhage, but it may rarely occur without massive bleeding. Nowadays, postpartum hypopituitarism is rarely seen owing to the current improvement in obstetric care and availability of rapid blood transfusion. Here we reported postpartum hypopituitarism in two siblings and discuss the possible etiology.

Case report: A 31-year-old female, with lassitude, agalactia, and loss of appetite for four months postpartum, visited our hospital in Nov, 2013. She had delivered her first baby by Caesarean section in Jul, 2013. During the delivery, the course was smooth and she was discharged without any apparent complications. But failure of postpartum lactation and failure to resume menses after delivery were mentioned. Her pituitary hormone function showed ACTH=4.36pg/ml, cortisol<1.0μg/dl, prolactin<2.2ng/ml, TSH<0.03μIU/mL, and FT4=3.23ng/dl, demonstrating that she had partial hypopituitarism with thyrotoxicosis. She was negative for anti-TSH-R Ab, anti-microsomal Ab, and anti-thyroglobulin Ab. Postpartum thyroiditis was suspected, and transient thyrotoxicosis was followed by secondary hypothyroidism with FT4 decreasing to 0.29ng/dl two months later. Interestingly, she has an elder sister who presented to our hospital two years later at the age of 37, with similar symptoms of agalactia and loss of appetite for two months postpartum. This was the second pregnancy of her elder sister, whose pituitary function also confirmed postpartum hypopituitarism, with ACTH=6.96pg/ml, cortisol<1.0μg/dl, prolactin<2.2ng/ml, TSH<0.03μIU/mL, and FT4=1.16ng/dl decreasing to <0.29ng/dl two months later. The pituitary MRI of the two siblings did not show any hemorrhagic sign or empty sella. Both of the siblings have been taking hormone replacements with glucocorticoids and thyroxine since diagnosis.

Discussion: The pathogenesis of postpartum hypopituitarism is not clearly understood. While the presence of anti-pituitary antibodies have been demonstrated suggesting an autoimmune process, one case report about two siblings has identified the Prophet of Pit-1 (PROP-1) gene defects. We reported the two siblings diagnosed with postpartum hypopituitarism, indicating that a genetic etiology exists and needs to be elucidated.