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A successful pregnant outcome in patient with Sheehan syndrome: a case report

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Summary

Sheehan syndrome, which is defined as hypopituitarism, is usually caused by massive postpartum hemorrhage. Hormone replacement therapy is recommended in patients with Sheehan syndrome. It is rare that pregnancy occurs in a patient with Sheehan syndrome, which is associated with high risk of maternal and fetal mortality. The authors report a 36-year-old twin pregnancy with Sheehan syndrome after the first pregnancy. She was treated with adequate management by obstetrician and endocrinologist cooperation during the pregnancy and delivery and had a successful outcome of this pregnancy. This case highlights the importance of proper management for the outcome of pregnancy in patients with Sheehan syndrome.

Key words: Sheehan syndrome; Postpartum hypopituitarism; Massive postpartum hemorrhage.

Introduction

Sheehan syndrome, which was first reported in 1937, is name for postpartum hypopituitarism caused by pituitary necrosis [1]. Massive postpartum hemorrhage, complicating by shock and disseminated intravascular coagulation (DIC), is one of causes of ischemic pituitary [2]. Due to hypopituitarism, pregnancy in patients with Sheehan syndrome is uncommon. In addition, the incidence of abortion and maternal and fetal mortality increase in patients with Sheehan syndrome [3]. The adequate management is crucial for pregnancy with Sheehan syndrome.

The authors present a successful pregnancy outcome of a patient with Sheehan syndrome. This case report was conducted in accordance with the appropriate clinical and experimental ethical guidelines and was approved by the Ethical Committee of the First Affiliated Hospital, Sun Yat-sen University. The case was reported with informed consent from the patient and her relatives.

Case Report

The pregnancy occurred in a 36-year-old woman with previous Sheehan syndrome, that presented to this hospital on November 14, 2015 for threatened labor. There was no remarkable abnormality in general physical examination. Previous medical records stated that the patient had had a stillbirth with vacuum extraction in 2012 at the age of 33 years, and delivery that was complicated by recessive prolapsed of umbilical cord. She went into shock after massive uterine hemorrhage (2,500 ml blood loss in two hours after delivery), complicated with DIC, and received 1,200 ml blood transfusions at the local hospital. She was diagnosed with Sheehan syndrome in May 2013 and received replacement therapy. She was on the treatment in the form of prednisolone 5.0 mg/day, thyroxine eight μg/day, and cyclic estrogen/progesterone combination. Spontaneous menstrual bleeding arose after this treatment. On investigations the gonadotropin concentrations following resumption of menstruation were FSH 7.07 IU/L (normal 4.0−13.0 IU/L), LH 4.10 IU/L (normal 1.0−18.0 IU/L), serum cortisol 4.90 μmol/L (normal 2.90−19.40 μmol/L), and ACTH cortisol level was 3.82 pmmol/L (normal 0−10.02 pmol/L).

The woman bacame pregnant via intrauterine insemination (IUI) on March 13, 2015. During pregnancy, the thyroxine dosage was increased to 25 μg/day and the prednisolone dosage was 10.0 mg/day (five mg po ante jentaculum; five mg po ante coenam) to maintain the normal free thyroxine (TSH, 0.809 mIU/L; T3, 4.059 pmol/L; T4, 14.056 pmol/L) and serum cortisol concentration (8 a.m. cortisol, 3.50 μmol/L; 4 p.m. cortisol, 9.90 μmol/L). At 37+4 weeks of gestation, she underwent cesarean section in view of precious pregnancy with multiple seriously complications. During the delivery she was given prednisolone 100 mg injection intravenous drip with intensive intraoperative monitoring. Oral prednisolone (20 mg per os 8 a.m.; 10 mg per os 4 p.m.) replaced the rejection in puerperium. A healthy live 3.27 kg male baby was delivered with an Apgar score of 10. The monitoring was continued postoperatively for 72 hours. Both infant and mother are well.

Discussion

Sheehan syndrome exhibits partial or complete hypopituitarism caused by ischemic pituitary necrosis due to obstetrical shock, accompanied by DIC following severe postpartum hemorrhage [4]. However, it is difficult to predict the possible onset of Sheehan syndrome in patients suffered from massive hemorrhage during delivery, while most cases had mild hypopituitarism and failed to be diagnosed for a long time [5]. A study of 60 patients with Sheehan syndrome described that the average time between the pre-

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vious obstetric event and diagnosis was 13 years [6]. Almost 75% of pituitary has been destroyed before diagnosis [7]. Recently, there is no adequate marker to predict the early diagnosis of Sheehan syndrome in patients with massive postpartum hemorrhage. In the present case, the patient had been diagnosed six months later and received proper management. Volume of hemorrhage, shock index (S.I.), DIC score, and empty sella have been considered as obstetrical factors in development of Sheehan syndrome [8]. Nevertheless, its pathogenesis is still unclear.

It is well known that pregnancy in patients with Sheehan syndrome is rare, secondary to the dysfunction of gonad and effect of sex hormone axis [3]. In clinics, ovulation induction which includes administration of hCG and GnRH and in vitro fertilization (IVF) could induce ovulation and pregnancy in patients with Sheehan syndrome. In this case, the patient underwent artificial menstrual cycle treatment and had the second pregnancy via IUI technique.

Although there are some reports of spontaneous pregnancy or IVF in patients with Sheehan syndrome, the pregnancy outcome is seldom descried. Pituitary hormone deficiency was demonstrated to increase the miscarriage rate in pregnancies [3]. Furthermore, previous study showed an increased risk of postpartum hemorrhage, still birth, transverse lie, and small for gestational age fetus (SGA) in pregnancies with Sheehan syndrome [9]. Hormone replacement therapy requires adjustment, closely followed by the endocrinologist due to the hormonal changes during pregnancy. The supplementation of cortisol and thyroxin should be increased, as well as the hepatic corticosteroid-binding globulin and thyroxin-binding globulin increases stimulated by placental estrogen during pregnancy [10]. In the present case, monitoring of hormone level was done each month and dosage of prednisolone had been adjusted by endocrinologist during the perioperative period. Both patient and infant were in a good state after cesarean section. As far as the delivery mode for pregnancies with Sheehan syndrome is concerned, selective cesarean section may prevent adverse neonatal outcome, including transient tachypnea of newborn (TTN), respiratory distress syndrome (RDS), and so on. The unpredictable endocrinal complications following a long-term vaginal delivery increase the risk of mortality; therefore, cesarean section was performed in this case.

The present case highlights that successful outcome can occur in pregnancy with Sheehan syndrome. In summary, early diagnosis and proper management impact the outcome of pregnancy with Sheehan syndrome. Strict prospective follow-up is necessary in patients with massive postpartum hemorrhage in order to identify the cases with Sheehan syndrome. Furthermore, hormone replacement therapy is required with careful adjustments made by the endocrinologist during pregnancy.

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