

CASE REPORT

Ovulation Induction, Pregnancy and Delivery in a Patient with Partial Hypopituitarism due to Lymphocystic Hypophysitis: A Case Report.

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Abstract

Lymphocystic hypophysitis is an unusual autoimmune disease that causes partial or total hypopituitarism and often associated with chronic anovulation. We report a case of ovulation induction, uneventful pregnancy and vaginal delivery in one of our patient with lymphocystic hypophysitis.

Key words: Lymphocystic hypophysitis, chronic anovulation and ovulation induction.

Introduction

The classical studies of Sheehan have shown that the pituitary gland is very vulnerable during pregnancy and puerperium.¹ This vulnerability may lead to partial or even total hypopituitarism. Unlike in hypopituitarism due to Sheehan's syndrome, that due to lymphocystic hypophysitis may or may not follow pregnancy, or overt postpartum haemorrhage.^{2,3}

Lymphocystic hypophysitis (LH) is a rare pituitary gland inflammatory disease of suspected autoimmune aetiology.^{2,3,4,5,6,7,8,9} Most of the cases occur in young women during pregnancy or the first postpartum year.^{3,4,5,6} It presents with varying degrees of hypopituitarism and often in association with autoimmune diseases. In this syndrome, in the early stage a mass lesion is often seen on the MRI or CT scan which when biopsied consist of lymphocystic infiltration.^{2,3} The mass being inflammatory in origin later disappears. The clinical feature is that of panhypopituitarism or in most cases partial hypopituitarism or isolated deficiency.^{2,3,6,7} This includes amenorrhoea, infertility, weakness, cold intolerance, fever, confusion, nausea and vomiting, diarrhoea, hypotension and hyperkalemia.

We report a case of a Nigerian woman with partial hypopituitarism secondary to LH in whom a successful ovulation induction was followed by an uneventful pregnancy of a healthy baby following misoprostol induction of labour at term.

Case

A 39-year-old lady was referred to our infertility clinic from the hospital's medical unit for management of infertility. She was on treatment for hypothyroidism and cortisol deficiency secondary to LH. She was a multiparae, Para 2+1 all females. Her last delivery 7 years ago was uneventful. She has suffered LH during her last delivery. There was no history of postpartum haemorrhage. She breast-fed her baby for six months. She had repeated admissions for unexplained weakness, fatigue, fever, nausea and vomiting and diarrhoea since her last delivery. Because of this her senior sister residing in London invited her for further evaluation in London, where she was diagnosed to have pituitary hypothyroidism and cortisol deficiency due to lymphocystic hypophysitis. She was placed on replacement therapy with hydrocortisone and thyroxine and following, which she recovered. Her follow up since

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she came back has been in our medical unit from where she was referred to us.

On evaluation at the infertility clinic, she was on hydrocortisone (20mg daily in divided doses) and L – thyroxine 0.1mg daily. Clinical examination, routine serum biochemistry was normal. Endocrinological evaluation showed ACTH, TSH deficiency, low FSH, LH and progesterone with normal prolactin level. Semen analysis, tubal patency test were essentially normal. Folliculometry and 21day progesterone assay confirmed anovulation as the possible cause of her infertility. Ovulation induction was initiated with pergonal (human follicle stimulating hormone 75IU/ human luteinising hormone 75IU) and choriegon(human chorionic gonadotrophin 5,000IU). Luteal phase support was given with micronised progesterone 300mg twice daily. Therapy was successful in the third cycle. Depot progesterone 250mg IM was given weekly until the 13th week. Early scan confirmed a single intrauterine pregnancy at 6weeks gestation. She was maintained on the prepregnancy doses of thyroxine and hydrocortisone because hormone assay at 12week showed normal free thyroxine and cortisol levels.

During the antenatal period she was seen fortnightly until 34weeks and thereafter one weekly till delivery. Antenatal screening was performed according to the hospital protocol. Ultrasound scan, serum biochemistry, free thyroxine hormones were repeated at 28, 32 and 36 weeks and they remained normal. She was also asked to keep fetal movement charts from 28weeks, which was inspected at every visit.

At 38 weeks, ultra sound scan results showed a mild Oligohydramnios, non stress test showed a reactive fetus. Labour was thus induced with misoprostol 100 microgram. She progressed to full cervical dilatation after 8 hours in labour. She had a vacuum delivery of a live male baby birth weight 3.6 kg and Apgar score of 7 and 9 at the first and the fifth minutes. During the labour period she was on intravenous hydrocortisone 200mg stat and then 6hourly until delivery. She had bilateral tubal ligation by modified Pomeroy technique on the first day postpartum. Postpartum period was uneventful. She was referred back to the medical unit for continued care.

Comment

Lymphocytic hypophysitis is an uncommon disorder with a striking female predilection and is related to pregnancy in about 70% of affected women.^{2,3} The cause is unknown, but the finding of anti-pituitary antibodies in some patients and its association to other autoimmune diseases, mainly thyroiditis, suggests an immune origin.^{2,3,4} In our patient, the beginning of the

picture during pregnancy/puerperium is similar to other reported cases elsewhere.^{4,5,6,7}, but distinct from cases reported by Garcia-Miguel et al² and Van Havenberg et al⁸ occurring outside pregnancy.

Only one case of ovulation induction in a woman with LH had been reported, to the best of our knowledge². Several authors advise the addition of GH to classical treatment with hMG/hCG in “poor responders” or GH-deficient patients,^{2,9} but this is controversial. In this patient we used FSH/LH with HCG for ovulation induction. We did not add growth hormone because there was no evidence of growth hormone deficiency, more so we were able to achieve ovulation in the first cycle and pregnancy in the third cycle treatment was used.

The natural history of LH, as well as the influence of further pregnancies on it, is unclear^{2,4,6}. There is a close association between pregnancy and the initial occurrence of this disease, so the possibility of deterioration during subsequent pregnancy was a matter of concern to us, we thus counseled her on a permanent contraception. She consented and had bilateral tubal ligation by modified Pomeroy technique. The course of the pregnancy was normal, as could be expected, because replacement therapy with close monitoring was provided^{2,4,5,6,7}. Delivery of the baby was decided on the 38th week, because of mild oligohydramnios.

Labor induction with misoprostol was chosen because of its efficacy and safety and more so oxytocin should be avoided in patients like ours because of the risk of water intoxication.^{10,11} A strict fluid balance, avoiding water generating solutions, was necessary, dextrose in saline being the choice.¹⁰

Corticosteroid supplementation should be increased for any patient being treated for chronic hypoadrenocorticism who undergoes a surgical procedure to prevent possible adrenal crisis.^{2,3} The degree of perioperative stress determines the dose and duration of therapy. Recommendations for perioperative steroid supplementation vary from author to author.^{2,4,5} For minor surgery IV hydrocortisone 25 mg preoperatively and 50 mg intraoperatively is frequently recommended to supplement their daily prescription. For major surgery hydrocortisone 25 mg preoperatively and 100 mg intraoperatively or dexamethasone 0.75 mg preoperatively and 3 mg intraoperatively are recommended.^{12,13} In this patient, we used IV hydrocortisone 100 mg statum then six-hourly. Oral treatment was resumed after delivery.

Our patient had partial panhypopituitarism evidenced by the low gonadotrophin and normal growth hormone

levels. Pregestational hormone replacement therapy was maintained, and she did not need adjustment in L-thyroxine and hydrocortisone level during pregnancy because repeated investigations showed normal hormone values.

In conclusion, we reported a case of ovulation induction in a woman with chronic anovulation, cortisol deficiency and hypothyroidism secondary to LH. The patient ran a course similar to that of other reported cases of hypopituitarism due to LH. Therefore a history of LH should not be considered a contraindication to pregnancy.

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