Spontaneous successful pregnancy in posthypophysectomy hypopituitarism: A rare case report

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ABSTRACT

Pregnancy in patients with pan-hypopituitarism following surgery of pituitary adenoma is rare and considered high risk. Hormonal dysfunction in these patients involves more than one axis (gonadotrophic, thyroidal, and adrenal). However, advance in infertility treatment have led to the increased pregnancy rate in hypopituitarism women. We present a case of nonfunctioning pituitary macroadenoma, who after pituitary surgery (hypophysectomy) developed hypopituitarism followed by multiple tuberculoma brain with hydrocephalus with arachnoiditis. She conceived spontaneously after 9 years of pituitary surgery and carried her pregnancy to the term. Elective caesarean section was done at 38 weeks and both infant and mother are well. The case highlights the rarity of the phenomenon and the safe outcome of the pregnancy with proper replacement.

KEY WORDS: Hypopituitarism, ovulation induction, pregnancy, pituitary macroadenoma, surgery

INTRODUCTION

Pregnancy in patients with pan-hypopituitarism is rare and associated with high risk of abortions, fetal, and maternal mortality. There also have been some reports of spontaneous pregnancy in patients with hypopituitarism, but there is little data on the pregnancy outcome.

CASE REPORT

A 33-year female with previous history of a nonfunctioning pituitary macroadenoma at the age of 24 years presented to our OPD for primary infertility treatment. On provoking and as per previous records she had headache and progressive loss of vision in both eyes for 4 months at that time of illness. Records of clinical examination at that time revealed temporal visual field defect with optic disc pallor and hemiaopia on the left side. The rest of the examination was unremarkable. Hormonal analysis was suggestive of hypothyroidism (TSH 0.097 μIU/ml) with appropriately normal gonadotrophins (LH, 0.39 mIU/ml and FSH, 0.97 mIU/ml), serum 8 AM cortisol (5.57 μg/dl) and normal prolactin (7.27 ng/ml) levels. Contrast-enhanced MRI and CT scan of brain revealed 2.9 × 2.5 cm pituitary mass lesion extending from intrasellar to suprasellar region abutting the optic chiasm [Figure 1a]. A diagnosis of nonfunctioning pituitary (NFP) macroadenoma was made. The patient subsequently subjected to transsphenoidal microsurgical tumor excision. Postoperatively she was followed with clinical, biochemical assessment and imaging. There was no evidence of hyperprolactinemia, diabetes insipidus, or any residual sellar tumor on MRI.

Bilateral vision started improving slowly and slowly and she developed central hypothyroidism, cushingoid features, and secondary amenorrhoea due to hypogonadotrophic hypogonadism. She was on replacement therapy with L-thyroxine 100 μg/day and prednisolone 7.5 mg/day. After 4 months, patient had one episode of seizure with loss of consciousness. On further examination and investigation, MRI multiple tuberculoma brain with hydrocephalous

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was found [Figure 1b]. Antiepileptic (carbamazepine 300 mg) and antitubercular (5-drug, SHREZ regimen) treatment for one year was started. For hydrocephalus the ventriculo-peritoneal shunt surgery was done at our institute. Patient was improved after this surgery and treatment. After a year of this she was started on cyclical estrogen (0.625 mg) and medroxy-progesterone (10 mg) therapy for withdrawal bleeding, which continued for 6 years. After this treatment patient had spontaneous menstrual bleeding. After 6 years, on investigations the gonadotropin concentrations following resumption of menstruation were FSH- 3.49 IU/L (normal 2.5–10.2 IU/L), LH- 2.31 IU/L (normal 1.9–12.5 IU/L) and the basal cortisol level was 405 nmol/l (normal 138–690 nmol/l) and 60 min post ACTH cortisol level, (also known as Synacthen test, normally double of basal values) was 824 nmol/l after withdrawing prednisolone, so prednisolone was stopped.

Patient was in our follow up. After a year of marriage patient conceived spontaneously, she was on antiepileptics. She received progestational support with micronized progesterone (Susten) and folic acid. In the first trimester, the thyroxin dosage was increased to 150 μg/day and later in the third trimester to 225 μg/day to maintain a high normal free Thyroxine concentration (greater than 1.5 ng/dl). The antenatal period was unremarkable. At 38 weeks of gestation, she had undergone elective caesarean section under general anesthesia in view of precious pregnancy with multiple neurological illnesses. During delivery she was given usual doses of thyroxin with intensive intraoperative monitoring. A healthy alive 2.9 kg female baby was delivered with an Apgar score of 10. The monitoring was continued postoperatively for 72 h. Both infant and mother are well, one month postdelivery and baby is on breast feeding.

DISCUSSION

Pregnancy in patients with underlying hypogonadotrophic hypogonadism is uncommon.[1] There also have been some reports of spontaneous pregnancy in patients with hypopituitarism, but there is little data on the pregnancy outcome.[2] Conception is reported in these patients occurring either spontaneously or with ovulation induction, but carrying successfully through to term is difficult, especially in the later condition. [3,4] There is increased risk of postpartum hemorrhage, transverse lie and small for gestational age fetus due to uterine dysfunction caused by hormone deficiency. A study reported live births in 61%, miscarriage in 28% and mid-trimester intrauterine death rate in 11% in the 18 patients.[5]

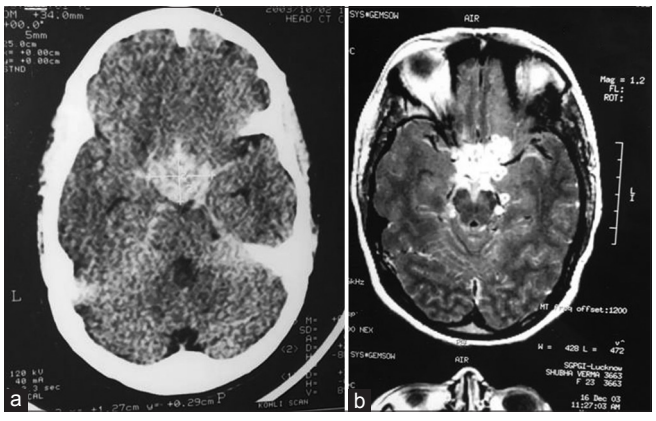

Figure 1: (a) CT scan brain showing pituitary mass. (b) MRI brain showing Tuberculoma postoperatively

After transsphenoidal adenomectomy, new unplanned hypopituitarism occurs in approximately 5% of patients, whereas improved hormonal function occurs in 50% of patients. The likelihood of new hormonal loss or recovery appears to depend on several factors. New hypopituitarism occurs most commonly in patients with tumors larger than 20 mm in size, whereas hormonal recovery is most likely to occur in younger, nonhypertensive patients and those without an intraoperative cerebrospinal fluid leak.[6,7]

In our case there was gonadal, adrenal, and thyroid failure following surgery for a pituitary macroadenoma. Amenorrhea was of pituitary origin because patient had withdrawal bleeding with estrogen and progesterone (EP) combination, but not with progesterone alone. Pregnancy in patients with panhypopitutarism can occur spontaneously, possibly due to periodic release of small quantities of gonadotrophins from any residual pituitary tissue or a combination of EP could assist in follicular growth. Pregnancies in women with panhypopituitarism are viewed as high risk. In our case, during the antenatal period, monitoring of thyroid functions and adjustments of the dosage of thyroxine was done to keep the FT4 levels in the high normal range and she needed 150–175 μg/day. She was lactating. The present case history highlights that spontaneous pregnancy can occur in patients with hypopituitarism and pregnancy can have successful outcome. Proper management is required during pregnancy and delivery in these patients.

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