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Case Report

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Case Report: Rare case of Secondary Idiopathic Pituitary Atrophy conceived after Intra Uterine Insemination (IUI)

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Abstract

Hypopituitarism or pitutary atrophy also known as empty sellaturcica is a disorder characterized by the deficiency of one or more of the hormones secreted by the pituitary gland. In this disorder patients may present with amenorrhea, poor pregnancy potential, infertility, and no production of milk after delivery. Here we present very rare case of pregnancy after intrauterine insemination in patient with secondary pituitary atrophy. The cause of her pituitary atrophy could not be exactly ascertained however it could be post-inflammatory.

Keywords: Intra Uterine Insemination; Pregnancy; Secondary Pituitary atrophy;

Introduction

Hypopituitarism or pitutary atrophy is a condition in which the pituitary gland shrinks or gets flattened. The pituitary gland is located in sellaturcica, compartment present at the base of the skull. It is not seen in MRI scan if pitutary gland is atrophied and looks like empty that is why it is also known as empty sellaturcica [1]. There are two types of sellaturcica. Primary empty sellaturcica occurs when there is a combination of (i) increased spinal fluid pressure and (ii) a defect in the diaphragmasellae, a membrane that sits on top of the pituitary. Primary empty sella is seen during pregnancy, obesity, and pseudotumorcerebri (a condition of increased spinal fluid pressure seen in obesity and associated with vision loss). **Secondary empty sella syndrome occurs** when the pituitary gland regresses after surgery to remove a pituitary tumor; radiation to treat a pituitary tumor; or a condition that damages the pituitary gland such as an old history of pituitary apoplexy that the patient was unaware of, hypophysitis, or neurosarcoidosis [2].

Case Report

A 32 years old female patient presented with primary infertility and irregular periods at Sahyadrisuperspeciality hospital IVF OPD at Karad, Maharashtra. She was married for two and a half years. Her husband is 35 years old. She is averagely built and nourished. Her growth and development were normal during her childhood. She attained menarche at 13 yrs age and her cycles were regular with moderate bleeding for 4-5 days every 20-30 days and cycles were painless. At the age of 17 yrs she gives history of high grade fever with cold and cough for 8 days which was treated by a local physician.

Following this episode of fever her menstrual cycle became irregular with bleeding for 2-3 days every 2-3 monthly. After 2-3 yrs she started getting menses only after taking medication. At age of 25 yrs she was diagnosed to have hypothyroidism an was put on medication. She got married at the age of 30 yrs and started

treatment for infertility after one year of marriage. She consulted a local gynecologist who advised her routine investigations.

The results were as follows:

- Serum cortisol : 2.59 ug/ml (Normal range 6.2 - 19.4)
- Insulin like growth factor (ILGF) : < 25 (116-358)
- Ultra TSH : 8.75
- Serum T3 : 1.4
- Serum T4 : 0.25
- Serum LH : 1.24
- Serum FSH : 3.39
- Serum Prolactin : 1.94
- Serum AMH : 3.66

She was put on thyronorm 62.5 mg once a day. She was treated for her infertility in the form of Letrozole 5mg once daily for 5 days for 6-7 months but no follicular growth was observed.

So, further investigations in the form of MRI scan of brain with contrast was advised which showed Empty Pituitary Sella. The pituitary gland is flattened against the floor of sella. The anterior lobe of pituitary is severely thinned out. The posterior lobe of pituitary appears small in size with normal signal intensity. No abnormal contrast enhancement seen. Represents moderate to severe atrophy of the pituitary gland.

When she came to our center she was married for 2 and half years. We repeated her routine hormone assays on day 2 of her periods which are as follows:

- Serum LH : 0.80
- Serum FSH : 2.80
- Serum AMH : 3.46
- Serum Prolactin : 1.89
- Serum TSH : 0.08

Transvaginal sonography: Hypoplastic uterus with thin endometrium. Bilateral ovaries appeared small with AFC of 1-2 on each side. We took a neurologist opinion who confirmed that there were no signs of raised ICP and fundii were normal. We diagnosed her as a case of pituitary atrophy and started her treatment for primary infertility. Husband investigations showed mild oligozoospermia. We started her stimulation with HMG (Human menopausal hormone) 75 IU X 3 doses and repeated her sonography on day 7 which showed one follicle of 9x9 mm in left ovary and endometrium of 4.5 mm. So we increased her dose of HMG to 150 IU for 3 days more. We did her sonography on day 10 which showed 2 follicles of 12x12mm and one follicle of 13x13 mm in right ovary and small follicles in left ovary with endometrial thickness of 6mm triple line

We increased the dose of HMG to 225 IU for 3 days more and rescanned her on day 13 which showed one follicle of 16.7x16mm and one follicle of 15x15mm on right side with ET of 7mm triple line. We stopped the injections and rescanned her on day 15 which showed two dominant follicles of 20x20mm on right side and ET of 8.5 mm. We triggered her ovulation with r.hcg (ovitrelle) did pre and post ovulatory IUI. We put her on estrogen and progesterone luteal support. We did her urine pregnancy test after 15 days which was positive. We continued her estrogen progesterone support till 12 weeks of pregnancy. She is continuing her pregnancy and now in her 28 weeks of gestation.

Discussion

Hypopituitarism or pitutary atrophy also known as empty sellaturcica. In this disorder patients may present with amenorrhea, poor pregnancy potential, infertility, and no production of milk after delivery. Achieving pregnancy in such patients can be difficult and very few cases of successful pregnancy have been reported. Here we present very rare case of pregnancy after intrauterine insemination in patient with secondary pituitary atrophy. Ssecondary EmptySelaSyndrome is due to destruction of the pituitary gland have symptoms that reflect the loss of pituitary functions, such as the ceasing of menstrual periods, infertility, fatigue, and intolerance to stress and infection [1].

Usually, the hormonal profile is normal in patients with empty sella. However, mild hyper-prolactinaemia (usually occurs in approximately 15% of patients [3].

Complications:

a) In view of the increased intracranial pressure and traction over the optic chiasma caused by the postsurgical adhesions, visual field defects are known to occur and often require treatment.

b) Cerebrospinal fluid rhinorrhoea can occur presenting as a non-traumatic and persistent nasal discharge.

c) Long standing increase in the intrasellar pressure can also lead to pituitary dysfunction, which needs to be corrected with appropriate hormone replacement therapy.

Total absence of diaphragmasella has been reported to occur in 20.5 % of normal subjects [4]. Barkan AL (1989) reported the cases of two women with anterior hypopituitarism after obstetrical catastrophies with blood loss and hypovolemia [5]. In case report by Dragojevic *et al* hMG injection was used for follicular development in a 38-year-old patient of Hypogonadotropic hypogonadism with empty sella syndrome that led to the formation of two follicles and after IUI, resulted in a successful singleton pregnancy and subsequent live birth [6]. As per Campo S et al case report, successful twin pregnancy was achieved in a 36-year-old patient of primary HH with empty sella syndrome after using recombinant FSH and LH [7].

Conclusion

This case report is a rare presentation of successful pregnancy achieved after Intra Uterine Insemination (IUI) in a patient with secondary Pituitary atrophy.

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