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A CASE OF SPONTANEOUS OVARIAN HYPERSTIMULATION SYNDROME AND PITUITARY HYPERPLASIA SECONDARY TO HYPOTHYROIDISM IN PUBERTAL GIRL

Obstetrics & Gynaecology

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ABSTRACT

ovarian hyperstimulation syndrome is a condition usually associated with ovulation induction. Rarely a non pregnant woman or woman not on ovulation induction treatment has been documented to develop spontaneous ovarian hyperstimulation syndrome. In this case study, we describe a female in the pubertal age range who had spontaneous ovarian hyperstimulation and pituitary hyperplasia due to primary hypothyroidism, which improved with thyroid replacement therapy.

KEYWORDS

Spontaneous ovarian hyperstimulation, pituitary hyperplasia, puberty and hypothyroidism.

INTRODUCTION

Ovulation induction leads to the iatrogenic condition known as ovarian hyperstimulation syndrome (OHSS). Following high levels of HCG (normal pregnancy), hypothyroidism, and FSH mutation, spontaneous OHSS occurs . It is uncommon for females in their pubescent years to have hypothyroidism and OHSS due to pituitary hyperplasia. Growth arrest or premature puberty are used to make the clinical diagnosis. The signs and symptoms include pericardial effusion or pleural effusion along with numerous cysts of various sizes and bilateral symmetric enlargement of the ovaries. (1)

Pubertal age group girls present with enlarged multicystic ovaries, ascites, pleural effusion, and occasionally even elevated CA 125 values, leading the clinician to suspect ovarian cancer. In this case study, we describe the common imaging findings that may be utilized to establish the benign nature of this illness and prevent needless surgical morbidity. The inability to detect primary hypothyroidism as a potential cause of ovarian cysts might result in an unintentional oophorectomy. Primary hypothyroidism is an endocrine disorder that is extremely common around the world. (2, 3)

The research suggests that cysts may regress spontaneously with normalization of thyroid- stimulating hormone (TSH) and free thyroxine (T4) levels (4-6). This case report covers a pubescent girl's unusual clinical history. This case report emphasizes that all patients with spontaneous OHSS should be tested for hypothyroidism, as it can occur in non-pregnant women who are not on ovulation induction treatment.

Case Report

A 12-year-old pubertal girl presented with abdominal discomfort and vomiting for four days. Her face was puffy and she had coarse skin, tubular breasts, lower abdominal fullness, short stature, class II obesity, calf muscle hypertrophy. She was pale and her feet were edematous. She was moderately mental retarded. [figure: 1] Mother had a family history of pancreatic neuroendocrine tumour, which was surgically removed. She was hospitalized for further evaluation.

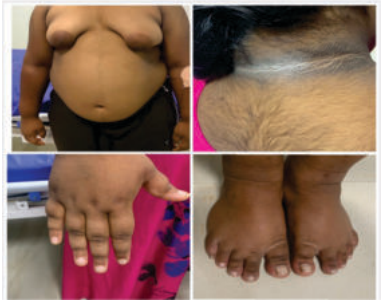

Figure: 1 Appearance Of The Patient In The Present Study

She was anemic, with a hemoglobin of 5.2 gm/dl (normal value 12-15 gm/dl), an increased TSH level of 608.90 micro IU (normal value 0.51 -4.30), and a low free thyroxine (T4) level of 0.13 ng/dl (normal range 0.98-1.63), indicating primary hypothyroidism.

A right ovary measuring 12.2x6.9x8.1 cm (TRXAPXCC) with a capacity of 360cc and a left ovary measuring 6.1cmx3.7cmx6.1cm (TRXAPXCC) with a volume of 88cc with various cystic alterations (biggest 6x6.5 cm), thin internal septations, and normal vascularity was seen on ultrasound abdomen and pelvis. [Figure: 2]

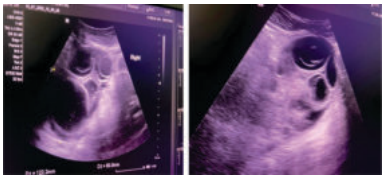

Figure: 2 Ultrasound Image Of The Study Patient

A 12-year-old girl was diagnosed with spontaneous OHSS after complaining of abdominal discomfort and vomiting. The abdominal ultrasound indicated bilaterally enlarged ovaries with several enlarged cysts and thin septations. The right ovary was 12.2x6.9x8.1 cm (TRXAPXCC), with a capacity of 360 cc, and the left ovary was 6.1cmx3.7cmx6.1cm (TRXAPXCC), with a volume of 88 cc with several cysts, the biggest measuring 6x6.5 cm.The ovaries in the pelvis seem substantially enlarged on MRI, with many thick-walled peripheral follicles of different sizes, indicating ovarian hyperstimulation syndrome.

MRI brain- homogenous T2 isointense pituitary mass lesion measuring 1.42x0.95x1.12cm (CCXAPXTR) with suprasellar extension.

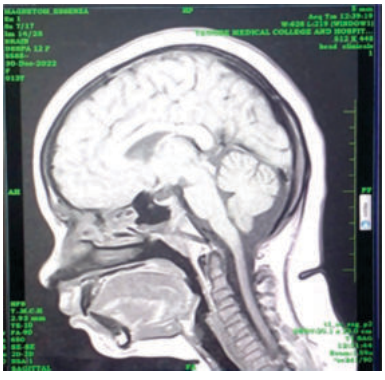

Figure 3: MRI Brain Of The Patient

An echocardiogram indicated a moderate pericardial effusion with a 65% ejection percentage. [figure: 4]

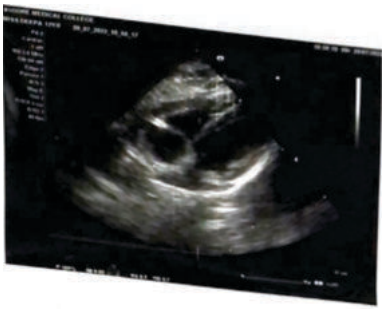

Figure: 4 Echocardiogram Image

Based on the radiological findings, the patient was evaluated further for ovarian malignancies. Alfa fetoprotein and CA-125 were normal. Hormonal investigations showed raised estradiol measuring 169.2 pg/ml (normal value <56pg/ml), low LH of value 0.451 mIU/ml (normal value 2.4-12.6) , normal FSH and beta HCG, serum prolactin of 101.80 ng/ml (normally less than 25 ng/ml).

OHSS, A diagnosis of spontaneous ovarian hyperstimulation syndrome and pituitary hyperplasia owing to hypothyroidism is determined based on the clinical, radiographic, and laboratory findings. Patient was started on Thyroxine 100 mcg/d and was followed up. With the help of two packed cell transfusions, anemia was corrected . patient was followed up for further management.

Repeated thyroid function tests after 4 weeks revealed a decrease in TSH to 6.07 mIU/ml and an increase in free T4 to 1.8 ng/dl. After two months of thyroxine replacement, TSH and free T4 returned to normal levels. Based on the hormonal readings, the thyroxine dose was gradually tapered , and the patient is currently receiving ongoing care.

Following four months of thyroxine replacement, an ultrasound of the pelvis revealed a decrease in the bilateral ovarian volume.

Right ovary measures about 4.9x5.2x3.1 cm (TRXAPXCC), a volume of 42cc and left ovary measures about 5.24.3x2.7cm (TRXAPXCC), volume of 32.6cc. Her abdominal distension resolved and her symptoms improved significantly.

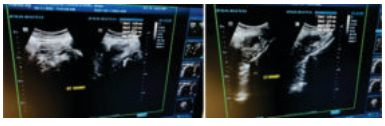

Figure 5: Ultrasound Pelvis After Four Months

MRI Brain was repeated after 6 months showed partial empty sella with no significant abnormality in the brain.

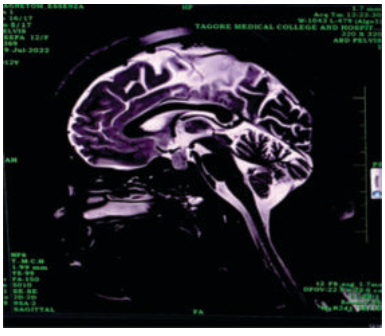

Figure 6: MRI Brain After Six Months

Patient was started on growth hormone and Leuprolide (gonadotropin releasing hormone agonist) given parenterally for the short stature and is in regular follow up now.

DISCUSSION

OHSS is an iatrogenic medical condition and is unique to stimulatory infertility treatment. (1) In this case, spontaneous OHSS is associated with primary hypothyroidism in a 12- year-old girl who attained menarche two months ago.

The risk factors for OHSS are young age, polycystic ovarian syndrome, asthenic habitus, luteal supplementation of hCG, protocols with GnRH, high level of serum estradiol and multiple follicles (>20 follicles), increased renin and angiotensin factors. The main pathogenesis is the increased vascular permeability leading to a fluid shift from intravascular to extravascular space.

Spontaneous OHSS has been reported between 8-14 weeks of pregnancy and also with follicle-stimulating hormone-producing pituitary adenoma. (7) Studies have shown its association with elevated human chorionic gonadotropin (hCG) seen in multiple pregnancies, polycystic ovarian disease, hydatiform mole and elevated Thyroid stimulating hormone (TSH) in hypothyroidism. (8, 9) hCG, TSH, FSH and LH belong to a family of glycoproteins and have two subunits- a common alpha subunit and a beta subunit specific to each molecule. Hcg and LH bind to LH receptors, FSH and TSH bind to FSH and TSH receptors respectively. (9)

Spontaneous OHSS is classified into three types based on clinical presentation and FSH receptor mutation. (10)

- Type I – related to FSH receptor mutation
- Type II – related to spontaneous OHSS secondary t high levels of hCG
- Type III – related to hypothyroidism

The exact mechanism by which OHSS occurs secondary to hypothyroidism is not clearly understood. A study by Rotmensch and Scommegna explained that preferential formation of estriol via the 16-hydroxylation pathway instead of 2- hydroxylation that is demonstrated in hypothyroid patients. Excessive gonadotropins release due to decreased feedback regulation caused by substituition of estradiol by estriol would result in excessive ovaraian stimulation. (11)

They can be classified as mild, moderate and severe. Mild characterized by uncomplicated ovarian enlargement with multiple cysts, moderate by ascites and severe by pericardial or pleural effusion, hemoconcentration, thrombosis, oilguria. The symptoms are due to ovarian enlargement and fragility, extravascular fluid accumulation and intravascular volume depletion. This leads to hypo albuminemia, hemoconcentration, electrolyte imbalance, decreased renal perfusion, oliguria, ascites and pleural effusion. This may cause significant mortality and morbidity from thrombosis, renal, liver and respiratory failure (ARDS). (11)

Imaging by US, CT, MRI reveal bilateral enlarged ovaries with multiple cysts, ascites and pleural effusion. Ovarian torsion may complicate to OHSS. PCOS should be ruled out.

Mild to moderate forms of OHSS are treated with analgesics and antiemectics. Severe form to be treated with hydration therapy and treating the underlying cause. In this case spontaneous OHSS was due to hypothyroidism treated with thyroxine.

CONCLUSION

This case report emphasizes that all patients with spontaneous OHSS should be tested for hypothyroidism, as it can occur in non-pregnant women who are not on ovulation induction treatment. All individuals with spontaneous OHSS should be evaluated for hypothyroidism, since this might be a cause of this unusual condition. Appropriate diagnosis at the appropriate time can assist physicians provide rapid care while also preventing subsequent issues.

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