Case Report

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Spontaneous ovarian hyperstimulation syndrome and pituitary macroadenoma associated with primary hypothyroidism

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Abstract

Ovarian hyperstimulation syndrome is associated with ovulation induction usually with gonadotropins and is rarely seen in pregnant women. Very few cases of spontaneous ovarian hyperstimulation syndrome have been reported in a woman who is neither pregnant nor undergoing ovulation induction therapy. Here we report an unsual case of a young adolescence girl with spontaneous ovarian hyperstimulation syndrome and pituitary macroadenoma. Her condition was triggered by unrecognized primary hypothyroidism, which regressed after treatment with levothyroxine and dopamine receptor agonist. This case highlights the need for clinicians and radiologists to familiarize themselves with the clinical and imaging features detected in this rare medical condition. Such improved knowledge will help to avoid delays in diagnosis, progression to life threatening complications, and unnecessary surgery. *[J Assoc Clin Endocrinol Diabetol Bangladesh, January 2022; 1 (1): 27-30]*

Keywords: ovarian hyperstimulation syndrome, pituitary macroadenoma

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Introduction

Ovarian hyperstimulation syndrome (OHSS) is usually iatrogenic and is a potentially life-threatening complication of ovulation induction. Spontaneous OHSS might occur following high levels of human chorionic gonadotropin (HCG) in normal pregnancy, hypothyroidism, or FSH receptor mutation.[1] Ultrasonography facilitates diagnosis and monitoring of this syndrome. The association of ovarian hyperstimulation with pituitary enlargement, although established in children and prepubertal girls with primary hypothyroidism, is rare.[2] Here, we present a new case of primary hypothyroidism complicated by spontaneous OHSS and pituitary macroadenoma in a non-pregnant adolescence girl. We also present imaging evidence of regression of both the ovarian and pituitary masses following thyroid hormone replacement and dopamine receptor agonist therapy.

Case Report

A 13-Years-Old girl presented with weight gain, headache with occasional vomiting, abdominal pain and distension and menstrual irregularity in the form of oligomenorrhoea followed by menorrhagia for last one year. On examination she was short (height <3rd centile), had features of hypothyroidism, distended abdomen with mild lower abdominal tenderness. Her laboratory evaluation reveals mild anemia, primary hypothyroidism and Hyperprolactinemia [Table 1]. Transabdominal ultrasonography revealed massive, bilateral, multiseptate cystic ovarian masses with a

Table I: Hormonal changes before and after treatment

Name	Initial evaluation	After 5-months
TSH	>138 µIU/ml	0.35 µIU/ml
FT4	0.16 ng/dl	1.71 ng/dl
Anti-TPO antibody	>1000 U/ml	
Prolactin	134.56 ng/ml	0.30 ng/ml
Estradiol	4748.8 pg/ml	39.58 pg/ml
LH	0.01 mIU/ml	2.82 mIU/ml
FSH	5.91 mIU/ml	5.44 mIU/ml
CA-125	27.80 U/ml	
Alpha fetoprotein	4.3 ng/ml	

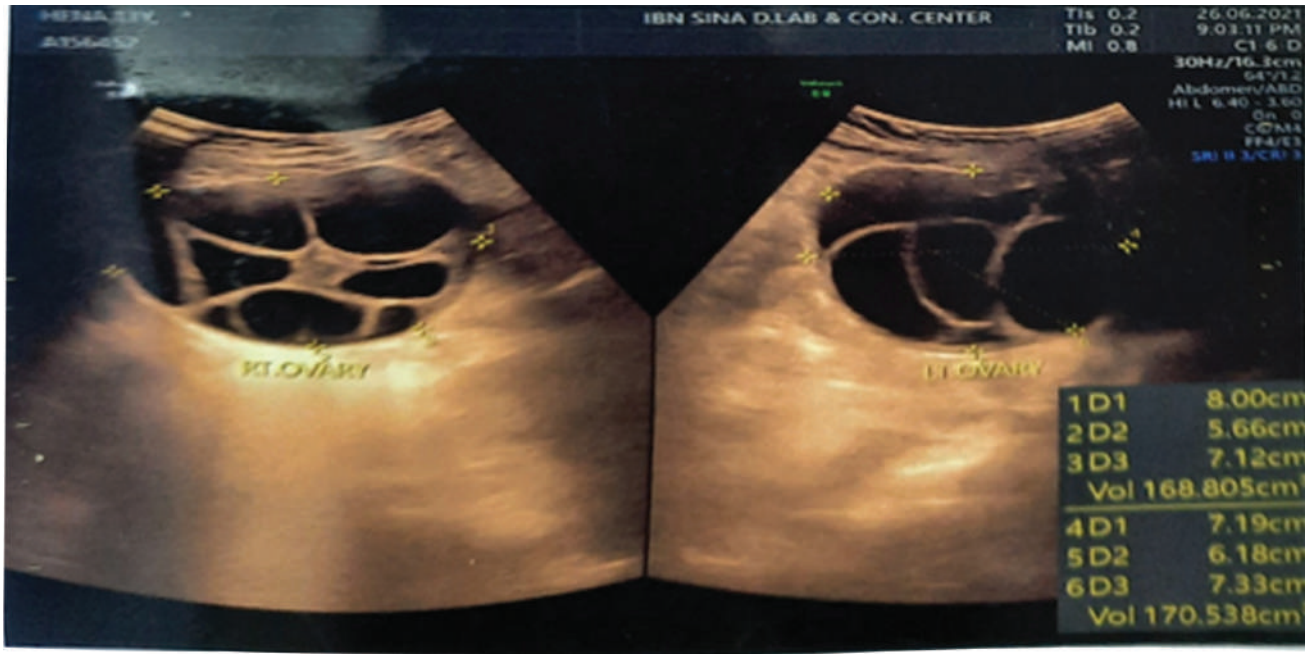

Figure-1: Ultrasonography shows soap-bubble or wheel-spoke appearance in both the ovaries

soap-bubble or wheel-spoke appearance in both the ovaries (Rt. ovary: 168 cm3, Lt. ovary: 170 cm3) [Figure 1]. Contrast enhanced MRI of sella and parasellar region revealed a 12 mm × 20 mm × 21 mm pituitary mass with suprasellar and parasellar extension. This lesion was heterogeneously isointense

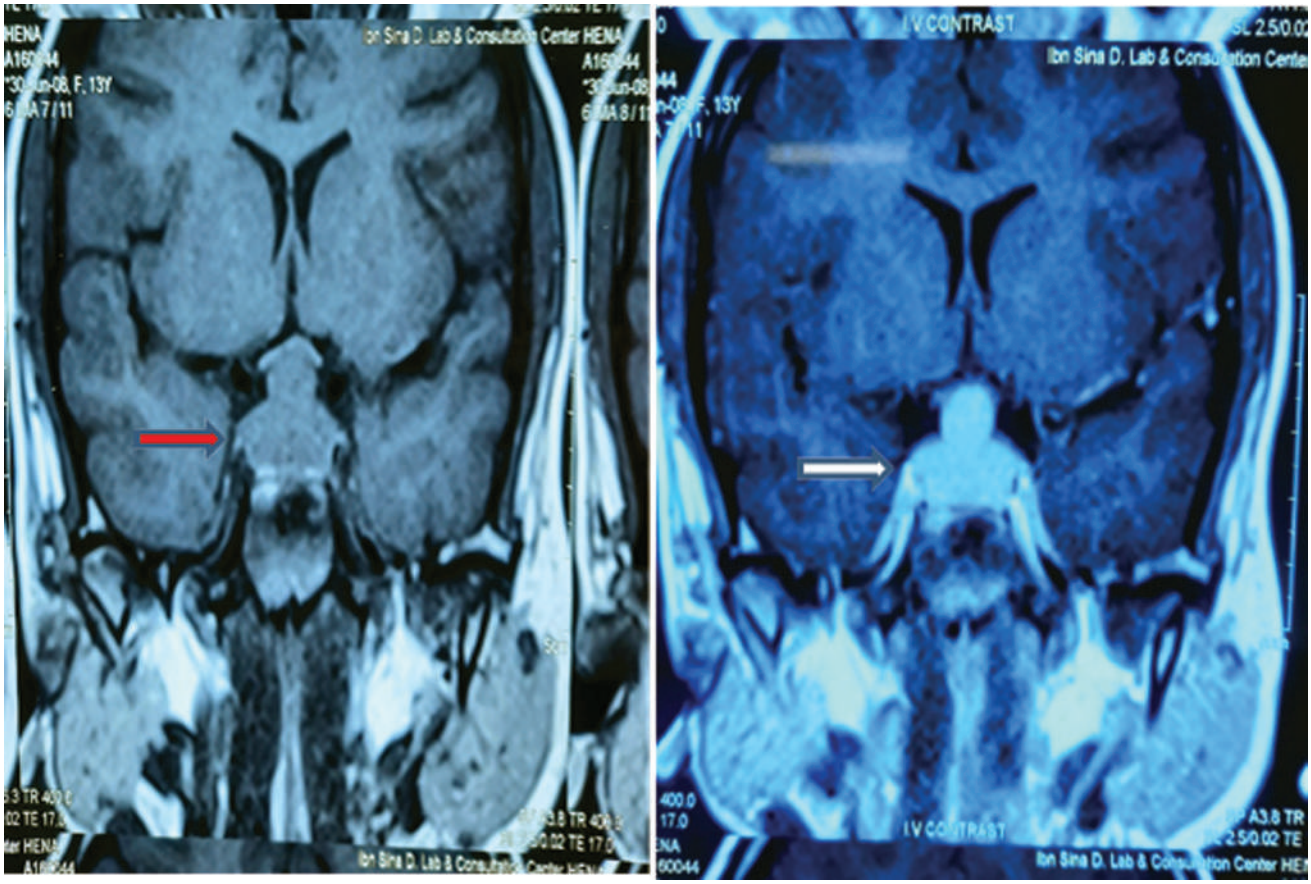

Figure-2: Pituitary MRI before (A) and after gadolinium contrast (B)

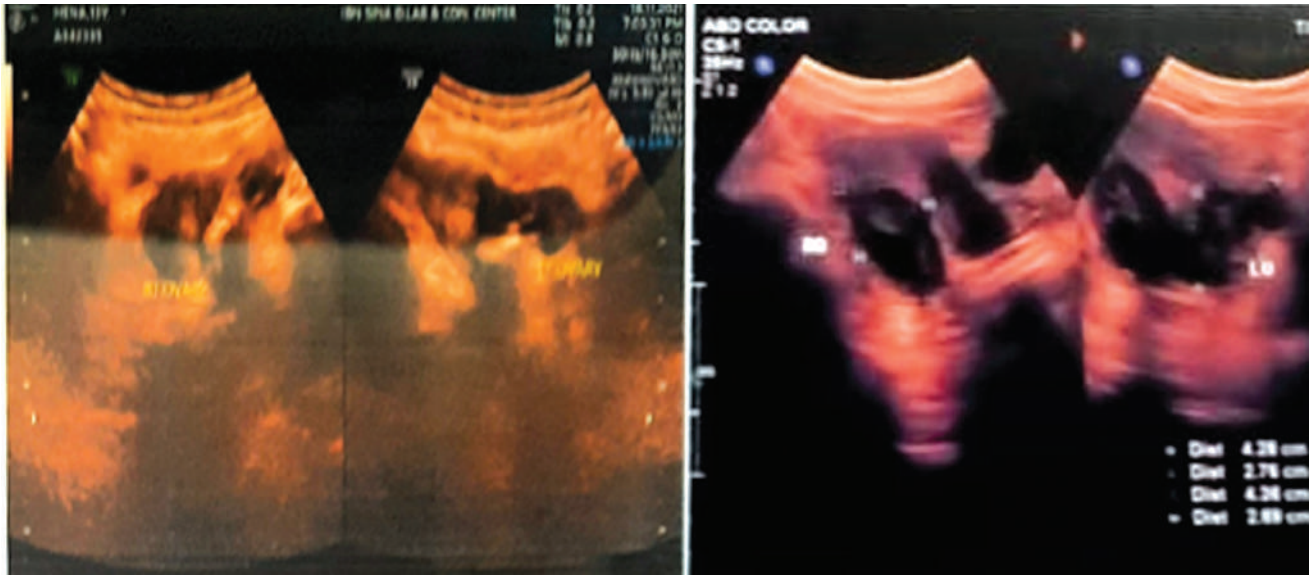

Figure-3A and B- Reduction of ovarian volume with time

on a T1 and T2-weighted image [Figure 2A]. Homogeneous enhancement of the mass was observed following gadolinium injection [Figure 2B]. The mass was reported to be suggestive of macroadenoma by the radiologist. Treatment started with levothyroxine and dopamine receptor agonist. On follow-up ultrasound, the size of the ovaries became significantly smaller 6 weeks after levothyroxine replacement and became normal with complete resolution of cysts after 05 months [Figs 3A and B]. Follow-up MRI 03 months later revealed regression of the pituitary tumor [Fig 4].

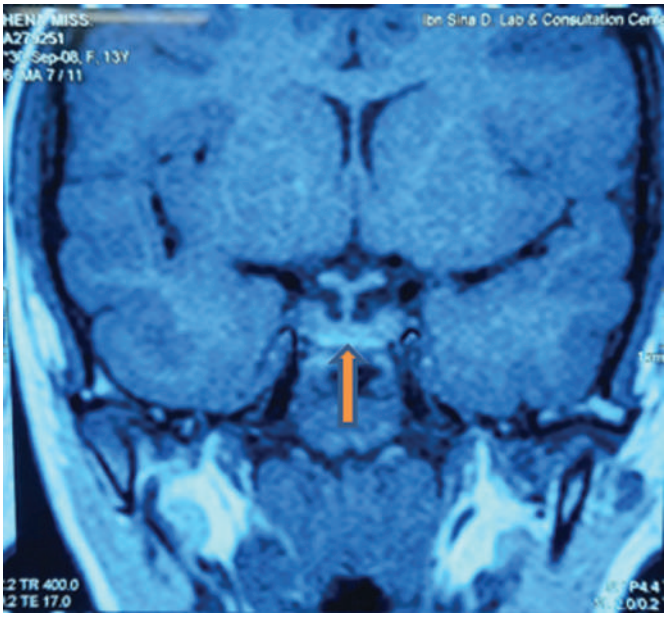

Figure-4: Regression of pituitary tumor

Hormonal status become normal after about 5 months [Table 1].

Discussion

Spontaneous OHSS is an uncommon clinical entity occurring due to non-iatrogenic aetiologies. In this case, it is called spontaneous OHSS, which has often been reported in association with pregnancy, FSH-secreting adenoma, or exceptionally TSH-secreting macroadenoma.[3] Only a few cases of hypothyroidism have been described in the literature.[4] The exact mechanism by which OHSS might occur in the context of hypothyroidism also remains unclear. Different mechanisms have been proposed: (1) TSH-mediated stimulation of the FSH receptor with[5] or without FSH receptor mutation,[6] taking into account that TSH has weak FSH activity and (2) the preferential formation of estriol in patients with hypothyroidism, which results in excessive gonadotropin release (estriol being a weaker suppressor of gonadotropin release than estradiol).[6] The clinical symptomatology can vary depending upon the severity of OHSS and occurs primarily due to extravascular accumulation of fluid. Hormonal evaluation in these patients reveals a markedly elevated TSH and raised anti- TPO-antibodies suggestive of autoimmune involvement. A wide array of investigations need to be performed to identify the aetiology of spontaneous OHSS. Ultrasonography in these cases reveals a classical soap bubble or spokes in wheel appearance of the ovarian masses.

In primary hypothyroidism, longstanding increases in thyrotropin-releasing hormone levels due to a lack of thyroid hormone induces both thyrotroph and lactotroph cell hyperplasia, resulting in TSH and prolactin over secretion. In the patient presented in this study contrast enhanced MRI of sella and parasellar region revealed a 12 mm × 20 mm × 21 mm pituitary mass with suprasellar and parasellar extension. This lesion was heterogeneously isointense on a T1 and T2-weighted image. Homogeneous enhancement of the mass was observed following gadolinium injection. So with the clinical presentations, USG findings and MRI findings we diagnosed her as a case of primary hypothyroidism complicated by spontaneous ovarian hyperstimulation syndrome and macroprolactinoma. After treatment with levothyroxine and dopamine agonist regression of ovarian volume and pituitary tumor was observed. This case highlights the need for clinicians
and radiologists to familiarize themselves with the clinical and imaging features detected in this rare medical condition. Such improved knowledge will help to avoid delays in diagnosis and prevent unnecessary surgery.

Acknowledgement
We are grateful to our patient and her parents for giving the consent to report the case.

Conflict Of Interest
The authors have no conflicts of interest to disclose

Financial Disclosure
The author(s) received no specific funding for this work.

Data Availability
Any inquiries regarding supporting data availability of this study should be directed to the corresponding author and are available from the corresponding author on reasonable request.

Ethics Approval and Consent to Participate
Written informed consent was obtained from study participant. All methods were performed in accordance with the relevant guidelines and regulations.

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