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Rare unusual case of spontaneous OHSS in a young unmarried girl

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Description

A 12-year-old female child attained menarche at 10 years of age with and presented with complaints of lower abdominal pain and irregular menses. The child was diagnosed with hypothyroidism at the age of 8 years and was put on thyroxine and is currently on 75 mcg thyroxine for the past 1 year with normal thyroid parameters. The child while being evaluated for pain abdoment and irregular periods underwent ultrasonography and MRI examination of the abdomen and pelvis which showed bilateral bulky adnexal with bilateral multiloculate T2/STIR hyperintense cystic lesions measuring ~6 x 7 x 8 cm on right side and ~7 x 4 x 9 cm on the left side (AP x TR xCC) [1]. These cysts do not restriction of diffucion on DWI or blooming on GRE ruling out presence of solid tissue or bleed. Uterus is normal with endometrial thickness of 8 mm. Urine pregnancy test was negative. The patients CA-125, LDH, AMH, progesterone, estrogen, testosterone and prolaction levels were all normal. Patients hCG, LH, FSH values were also within normal limits. Viral markers were also normal [2].

Discussion

Ovarian hyperstimulation syndrome (OHSS) is a severe disease with significant morbidity and mortality associated with it. It is usually caused by hyperstimulation of ovaries secondary to controlled pharmacological induction of ovaries for patients undergoing infertility treatment. Rarely it can occur in pregnancy or due to genetic component. Few associations are also seen with early pubertal development and primary hypothyroidism (Van Grumbach syndrome) [3,4]. Though the exact cause is not known but it occurs due to complex interplay of hormones at multiple levels and altered hormonal regulation through feedback loops. It is proposed that hypothyroidism caused increased TSH which in fact can stimulate FSH receptors. Similar stimulation is also anticipated at pituitary level causing increased TRH production stimulating GnRH receptor leading to ovarian enlargement. Usually the ovarian enlargement resolves with thyroid hormone replacement but in this case the ovarian hyperstimulation continued thereby suggesting a missing link in hormonal regulation. Extensive work up showed normal levels

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of hormones thereby further puzzling our pediatric and endocrine team. This is the first case to the best of our knowledge ever reported in literature in a young unmarried girl without pregnancy.



Figure 1: T2 haste axial images showing multiloculated bilateral adnexal cystic lesions.



Figure 2: STIR coronal images showing bilateral multiloculated adnexal lesions.



Figure 3: USG showing bilateral multiloculated adnexal lesions.

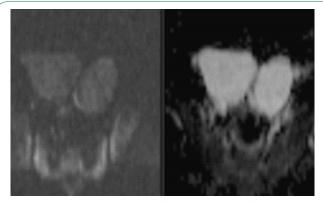


Figure 4: DWI images show that the lesions do not show restriction of diffusion.

Declarations

Conflicts of interest: Nil.

Informed consent: Informed consent taken from both the parents prior to submission for publication.

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