Spontaneous ovarian hyperstimulation syndrome revealing a pituitary macroadenoma

Sir,

Ovarian hyperstimulation syndrome is most commonly an iatrogenic complication of exogenous gonadotropin administration for ovulation induction. Its clinical presentation varies from abdominal pain and bloating, nausea, vomiting, and ovarian enlargement to possibly life-threatening conditions including renal failure, hypovolemic shock, adult respiratory distress syndrome, thromboembolism, and pericardial effusion.^[1]

Spontaneous ovarian hyperstimulation syndrome is a rare condition which has been described in association with pregnancy in patients with polycystic ovarian disease, severe primary hypothyroidism, and granulosa cell tumors.^[2]

A 36-year-old woman was referred to our department for hyperprolactinemia. She had irregular menses, and she has never been pregnant.

Previously, she presented to the Department of Gynecology with nausea, abdominal pain, and menometrorrhagia. Transvaginal ultrasound examination revealed bilateral multicystic enlarged ovaries (10 cm and 17 cm in diameter) without endometrial hyperplasia. A pelvic laparoscopy was performed, and multiple ovarian cysts were excised bilaterally. There was no evidence of malignancy.

Two years later, the patient presented with the same symptoms and a bilateral multicystic ovarian enlargement. She had no galactorrhea or headache.

On physical examination, she had a body weight of 70 kg, a body mass index of 20 kg/m² and a blood pressure of 90/60 mmHg. The abdomen was enlarged with the presence of a general tenderness with no sign of peritoneal irritation. The rest of the clinical examination was normal.

Laboratory tests demonstrated a negative pregnancy test, prolactin levels of 2842 mU/L (reference range 109-557 mU/L), thyroid-stimulating hormone level of 1.49 µIU/ml (reference range: 0.35–4.94 µIU/ml), free T4 level of 1.16 ng/dl (reference range 0.7–1.48 ng/dl), follicle-stimulating hormone (FSH) level of 6.8 mIU/ml (reference range 3.35–21.6 mIU/ml),

luteinizing hormone (LH) level of 0.6 mIU/ml (reference range 2.39–6.6 mIU/L), estradiol level of 68 pg/ml (reference range 25–75 pg/ml), adrenocorticotropic hormone level of 34 pg/ml (reference range 8–58 pg/ml), 08 h cortisol level of 448 nmol/L (reference range 101.2–535.7 nmol/L). Glycoprotein hormone α subunit was not available.

Magnetic resonance imaging (MRI) of the pituitary gland revealed a 21 mm \times 15 mm \times 17 mm pituitary adenoma with suprasellar extension [Figure 1]. Visual field examination was normal.

The diagnosis of macroprolactinoma was established and the patient was treated with bromocriptine. Three months later, laboratory tests revealed normal prolactin level. However, pituitary MRI showed no reduction in tumor size.

Thus, transsphenoidal pituitary surgery was performed with complete removal of the adenoma. Ultrasound evaluation after the operation revealed normal ovarian size and totally disappearance of the cysts.

In our patient, pituitary MRI was performed due to the detection of hyperprolactinemia. This exam revealed the presence of a pituitary macroadenoma. Thus, the diagnosis of macroprolactinoma was first established. However, the coexisting of a recurrent spontaneous ovarian hyperstimulation syndrome with a nonsuppressed FSH level and the totally disappearance of the cysts after surgical removal of the macroadenoma, are consistent with a mixed secretion of FSH.

In the literature, few cases of gonadotroph adenoma causing ovarian hyperstimulation have already been reported.^[3-5] High FSH levels resulted in the formation of multiple large ovarian cysts and significantly increased the production of estradiol.^[3]



Figure 1: Sagittal and coronal magnetic resonance imaging of the pituitary showing a 21 mm \times 15 mm \times 17 mm pituitary adenoma with suprasellar extension

Low LH and normal estradiol levels in our patient could be attributed to hyperprolactinemia that might have suppressed gonadotropin secretion or to insufficient LH production by the normal pituitary.

A similar case of ovarian hyperstimulation syndrome associated with FSH-secreting adenoma without elevated estradiol has been reported in literature.^[5]

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Conflicts of interest

There are no conflicts of interest.

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REFERENCES

- Halupczok J, Kluba-Szyszka A, Bidzinska-Speichert B, Knychalski B. Ovarian hyperstimulation caused by gonadotroph pituitary adenoma – Review. Adv Clin Exp Med 2015;24:695-703.
- 2. Roberts JE, Spandorfer S, Fasouliotis SJ, Lin K, Rosenwaks Z. Spontaneous ovarian hyperstimulation caused by a

follicle-stimulating hormone-secreting pituitary adenoma. Fertil Steril 2005;83:208-10.

- Pentz-Vidovíc I, Skoric T, Grubisic G, Korsíc M, Ivicevic-Bakulic T, Besenski N, *et al.* Evolution of clinical symptoms in a young woman with a recurrent gonadotroph adenoma causing ovarian hyperstimulation. Eur J Endocrinol 2000;143:607-14.
- Christin-Maitre S, Rongières-Bertrand C, Kottler ML, Lahlou N, Frydman R, Touraine P, *et al.* A spontaneous and severe hyperstimulation of the ovaries revealing a gonadotroph adenoma. J Clin Endocrinol Metab 1998;83:3450-3.
- Shimon I, Rubinek T, Bar-Hava I, Nass D, Hadani M, Amsterdam A, et al. Ovarian hyperstimulation without elevated serum estradiol associated with pure follicle-stimulating hormone-secreting pituitary adenoma. J Clin Endocrinol Metab 2001;86:3635-40.

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