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**Case Report**

# A rare case of ovarian hyperstimulation with torsion presenting with hypothyroidism

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## ABSTRACT

A thirteen years old female child, who was recently diagnosed with hypothyroidism and polycystic ovary and under thyroid supplements, presented with acute abdominal pain of one day duration. Examination showed pallor with rough skin, hypotension (70/40 mmHg) and distended abdomen. Abdomen was tense with tenderness and guarding. Urgent contrast CT abdomen showed bilateral polycystic ovaries with left ovarian torsion and hemo-peritoneum. Blood investigations showed anemia (6.0 g/dL) and TSH >100 microIU/ml. She was stabilized with Intravenous fluids, packed red blood cells and taken up for surgery. Laparoscopy confirmed the diagnosis and detorsion with deroofing of cysts was done. A post-operative diagnosis of Van Wyk Grumbach syndrome - Hypothyroidsm with ovarian hyper stimulation syndrome with hemorrhagic torsion of left ovary was made. She withstood procedure well and was stable. She was discharged to a local hospital after 2 days of surgery. She is on thyroid supplements and doing well.

**Keywords:** Van Wyk Grumbach syndrome, Hypothyroidism, Precocious puberty, Hyperprolactinemia, Laparoscopic deroofing and detorsion

## INTRODUCTION

Van Wyk-Grumbach syndrome (VWGS) is a rare complication of prolonged untreated juvenile hypothyroidism. The etiology has been thought to be related to complex interactions within the hypothalamic-pituitary axis and high levels of TSH acting on FSH receptors due to molecular similarities between the glycoprotein receptors of these two hormones, which share a common subunit.[1] Girls with Van Wyck-Grumbach syndrome can have varying degrees of pubertal development as well as multi-cystic ovaries, vaginal bleeding, galactorrhea and delayed bone age. Boys usually present with testicular enlargement without virilization.[2] Von wyk grumbach syndrome can be diagnosed by its salient clinical and radiological features.[3]

Few of the studies done before like Rastogi A et al, have reported a case of 10.7 years old female suspected of having van wyk grumbach syndrome and responded successfully to thyroxine therapy.[4] Branoswki et al have reported a 8 years old girl with long standing hypothyroidism who presented with features suggestive of von wyk grumbach syndrome.[5] Sneha et al, have reported a case of short stature diagnosed as having van wyk grumbach syndrome which responded well to thyroxine therapy.[6] Although the etiology of the disease remains unclear the treatment plan remains the same with patients responding well to thyroxine therapy. Patients respond well to thyroxine therapy with improvement in symptoms and resolution of symptoms.[6]

Hence, it can be concluded that early recognition of this entity is important as the early institution of thyroid

hormone replacement therapy can help in resolution of symptoms.

## CASE REPORT

A thirteen years old female child, who was recently evaluated in an outside hospital for menorrhagia, short stature and obesity (weight - 47 kg, for her age, normal weight is 45 kg), diagnosed with hypothyroidism and polycystic ovary and under thyroid supplements (Tab Thyronorm 50 microg), presented with complaints of abdominal pain for past one day and reduced food intake. She was found to have hyperprolactinemia during evaluation. Examination showed pale rough skin with low BP (70/40 mmHg), distended abdomen and a mass palpable with tenderness and guarding. External genitalia normal. She was immediately admitted for further management. Blood investigations and CT abdomen done on the same day. CT abdomen with contrast showed bulky ovaries with multiple cysts, moderate free fluid in the abdomen and pericardial effusion, suggestive of ovarian hyper-stimulation syndrome, bulky left ovary with absent contrast enhacement, cyst with hemorrhagic content and twisted left vascular pedicle, features suggestive of ovarian torsion and hyper-dense content within the fluid in the right lower abdomen, suggestive of hemo-peritoneum. Other structures normal. ECHO done showed pericardial effusion. Blood investigations showed anemia (Hemoglobin 6 g/dl) and TSH > 100 microIU/ml, other blood investigations turned out to be normal. She was stabilized and taken for surgery the next day after getting consent from her mother. Under general anesthesia, laparoscopic cyst deroofing with detorsion was performed. Around 500 ml of hemorrhagic fluid was aspirated, and as the ovary was large and bulky, de-torsion could not be done and hence de-roofing of the ovary done with harmonic scalpel and specimen sent for histopathological examination. Intra-operative blood transfusion, two units were given to the patient in view of low hemoglobin and a post-operative diagnosis of Van Wyk Grumbach syndrome - Hypothyroidsm with ovarian hyper stimulation syndrome with hemorrhagic torsion of left ovary was made. She withstood procedure well and was stable. She was started on antibiotics and analgesics. Endocrinologist opinion was taken and Tablet Thyronorm 100 microg (Thyroid supplements) was prescribed. She was discharged on post-operative day 2 and was doing well. She was advised to continue the thyroid supplements and review after one week and appropriate safety netting was done.

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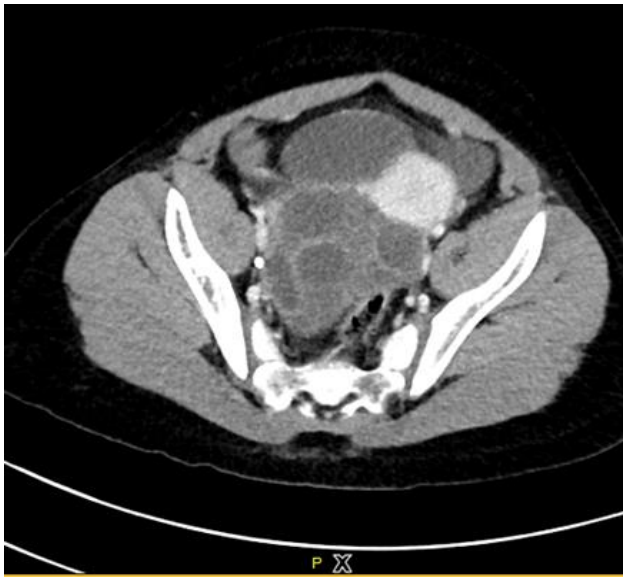

**Figure 1: CT with contrast AP view.**

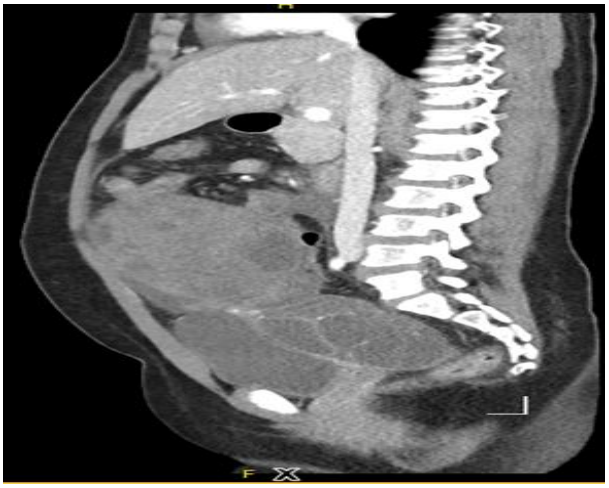

**Figure 2: CT with contrast lateral view.**

***Differential diagnosis***

Multiple cysts were seen in both the ovaries in a thirteen years old child, with menstrual irregularity and obesity, hence polycystic ovarian syndrome was thought as a differential diagnosis. But Polycystic ovarian syndrome has other features too like excessive hair growth, acne, loss of hair and insulin resistance which weren't seen in this child. Moreover, she has hypothyroidism which points more towards Van Syk Grumbach syndrome, and was treated accordingly.

***Treatment***

She was stabilized and taken for surgery the next day of admission after getting consent from her mother. Under general anesthesia, laparoscopic cyst deroofing with detorsion was performed. Around 500 ml of hemorrhagic fluid was aspirated, and as the ovary was large and bulky, de-torsion could not be done and hence de-roofing of the ovary done with harmonic scalpel and specimen sent for histopathological examination. Intra-operative blood transfusion, two units were given to the patient in view of low hemoglobin. She was started on antibiotics and analgesics to avoid infection and reduce pain. Endocrinologist opinion was taken and Tablet Thyronorm 100 microg (Thyroid supplements) prescribed.

***Outcome and follow-up***

She withstood the surgery well and was stable and started on antibiotics and analgesics. She was discharged on post operative day 2 and was doing well. She was advised to continue the thyroid supplements and review after one week and appropriate safety netting was done.

## DISCUSSION

Precocious puberty is a known, but rare complication of severe acquired juvenile hypothyroidism, and was first described by Kendle in 1905. It was not until 1960 when the term "Van Wyk and Grumbach syndrome" was coined. Van Wyk and Grumbach described a syndrome of precocious menstruation in juvenile hypothyroidism, with reversion to a pre-pubertal state after thy- roid replacement therapy. A clue to the diagnosis is the delayed bone age, because VWGS is the only form of precocious puberty in which the bone age is delayed.[7]

The most common cause of hypothyroidism in these patients is autoimmune thyroiditis. Pituitary hyperplasia, a common finding in VWGS, has been blamed on long standing thyrotrope hyperplasia in response to the decreased thyroid hormone. Hyperprolactinemia, usually found in laboratory data has two etiologies. Some postulate that the thyrotrope hyperplasia in the pituitary compresses the pituitary stalk disrupting hypothalamic inhibition of prolactin. TRH is also known to stimulate prolactin. When thyroid hormone is low, TRH increases lead to increased levels of prolactin.[8]

Anemia is not so uncommon in hypothyroidism and has been noted in several case reports of the Van Wyk–Grumbach syndrome. The proposed mechanism involves decreased red cell production in response to the reduced metabolic requirements for oxygen in tissues in hypothyroidism. In addition, the anemia may also be exaggerated by menorrhagia, dietary deficiency or pernicious anemia.[9]

Lastly, the multicystic ovaries may result from elevated levels of circulating gonadotropins acting on it. It is also possible that increased sensitivity of the ovaries to the circulating gonadotropins could result from the hypothyroid state directly or via increased prolactin. However, ovarian enlargement may be secondary to a myxedematous infiltration.[4]

## CONCLUSION

Hence, with this case report, it can be concluded that, early recognition of VWGS through salient clinical and radiological features is important, as early management through thyroid hormone replacement therapy can help in resolution of symptoms and preventing severe complications in future.

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