10.5005/jp-journals-10006-1370

CASE REPORT

Acute Abdomen, Primary Amenorrhea, Juvenile Hypothyroidism: A Rare Combination

[1]B Pushpalatha, [2]Radhika Potluri, [3]Vasantha Lakshmi, [4]Krishna G Seshadri, [5]PSNRS Sireesha

ABSTRACT

Ovarian cysts are common causes for gynecological surgery. Juvenile hypothyroidism, multiple large ovarian cysts causing acute abdomen and primary amenorrhea are infrequently reported and not widely recognized. We report a case of a 21-year-old lady who presented with juvenile hypothyroidism, primary amenorrhea, multiple cystic ovary and raised thyroid stimulating hormone (TSH), follicle stimulating hormone (FSH) and estradiol levels. After thyroid replacement, regression of TSH, FSH levels and cyst size were noted. Laparotomy done in view of torsion of ovary, histopathology (HPE) revealed multicystic ovary. This case was reported for its unusual presentation of hypothyroidism leading to torsion ovary.

Keywords: Juvenile hypothyroidism, Multicystic ovary, Primary amenorrhea.

How to cite this article: Pushpalatha B, Potluri R, Lakshmi V, Seshadri KG, Sireesha PSNRS. Acute Abdomen, Primary Amenorrhea, Juvenile Hypothyroidism: A Rare Combination. J South Asian Feder Obst Gynae 2015;7(3):236-239.

Source of support: Nil

Conflict of interest: None

Date of received: 19 September 2015

Date of acceptance: 20 November 2015

Date of publication: December 2015

INTRODUCTION

Massive ovarian enlargement, multicystic ovaries, ovarian hyperstimulation and amenorrhea have all been linked to hypothyroidism[1-4] Failure to recognize hypothyroidism as an etiology of ovarian cysts could lead to inadvertent oophorectomy. It has been recently shown that thyroid stimulating hormone (TSH) could interact with follicle stimulating hormone (FSH) receptors to elicit gonadal stimulation because through specificity spill over TSH could have an FSH and luteinizing hormone (LH) like effect.[5] Thyroid stimulating hormone could serve as a facilitating factor in the development of multiple follicular cysts.[4]

[1]Chief, [2]Postgraduate, [3]Associate Professor, [4]Head [5]Assistant Professor

[1-3, 5]Department of Obstetrics and Gynecology, Sri Ramachandra University, Chennai, Tamil Nadu, India

[4]Department of Endocrinology, Diabetes and Metabolism Sri Ramachandra University, Chennai, Tamil Nadu, India

Corresponding Author: Radhika Potluri, Postgraduate, Department of Obstetrics and Gynecology, Sri Ramachandra University Porur-600 116, Chennai, Tamil Nadu, India, Phone: 08056215309 09566191989, e-mail: radhi_1113@yahoo.co.in

CASE REPORT

A 21-year-old single woman presented with complaints of abdominal discomfort and distension for 1 month and primary amenorrhea. One month prior to admission, she noticed abdominal distention and vague abdominal pain. Patient has no history suggestive of anosmia, visual disturbances, headache, loss of weight appetite, childhood infections or prior chemo or radiotherapy. Mother attained menarche at 16 years. Medical and surgical history were otherwise unremarkable.

Physical examination revealed height—132 cm, weight—32.4 kg and BMI—18.6 kg/m^2. Skin was dry, no pallor, no bony deformities were noted. Thyroid was normal to palpation and not enlarged. She had poorly developed secondary sexual characters with breasts tanner stage 2 and no pubic or axillary hair (Tanner 1). There were no other dysmorphic features.

Systemic examination of cardiovascular, respiratory and central nervous system were normal. Abdominal examination revealed soft, 10 × 10 cm mass occupying the suprapubic and right iliac fossa, no ascites, no other organomegaly. External genitalia was normal with absent pubic hair and hymenal opening seen.

Initial investigations are summarized in Table 1. Hemogram, liver and renal function tests were normal. TSH—>150, FSH—67 μ/ml and estradiol—2230 pg/ml and markedly low LH—0.07 μ/ml and FT4 levels. CA 125 and other tumor markers like LDH, alpha fetoprotein and beta-hCG were normal. Karyotyping done showed 46, xx female karyotype USG pelvis revealed a mass in the right adnexa 12 × 8 cm with dense internal echoes

Table 1: Laboratory data before and after thyroxine therapy

Test	*Initial values*	*Repeat values after 3 weeks*
TSH	>150	14.78
FT4	0.27 ng/dl	1.87 ng/dl
FSH	67.07 μ/ml	<0.03 μ/ml
LH	<0.07 μ/ml	<0.07 μ/ml
Sr estradiol	2230 pg/ml	30.6 pg/ml
Tumor size in USG	12 × 8 cm	9 × 6 cm

(Fig. 1), minimal vascularity seen, uterus and left ovary were normal, minimal free fluid was seen. Computed tomography scan (Fig. 2) showed large multiloculated cyst lesion of size 13 × 8 × 6 cm seen in the right adnexa extending up to the lower abdomen. Multiple enhancing septa are seen, right ovary was not visualized separately and left ovary was normal. Minimal fluid seen in lower abdomen. Three hypodense lesions largest of size 1.4 × 1.2 cm seen in the right lobe of liver. A differential diagnosis of (1) cyst adenocarcinoma of right ovary with, (2) hemorrhagic metastasis in liver was made. ECG and Echo were normal X-ray of left wrist for bone age showed less than 15 years but greater than 11 years.

The patient was started on levothyroxine 125 mcg. She had vaginal bleeding 1 week after starting thyroxine treatment. Three weeks later, after treatment with thyroxine, there was drastic decline in TSH to 14.78, FSH to < 0.03 μml and Sr estradiol to 30.6 pg/ml (Table 1).

A repeat ultrasound done after 4 weeks showed decrease in tumor size 12 × 8 cm to 9 × 6 cm. Patient developed signs of acute abdomen with suspected torsion, taken up for laparotomy.

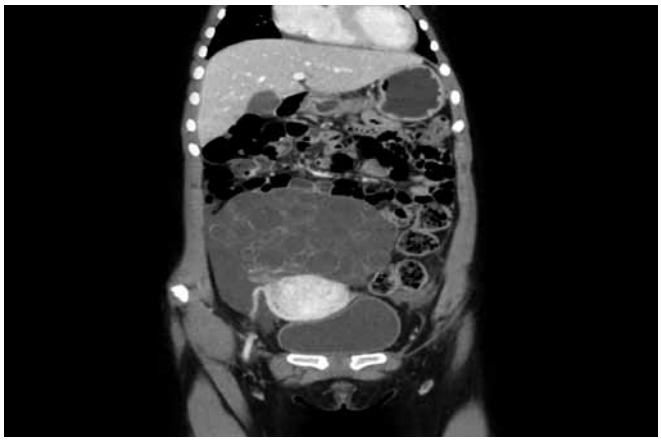

Fig. 1: Ultrasonography of abdomen right adnexa mass 12 × 8 cm with dense internal echoes and minimal vascularity

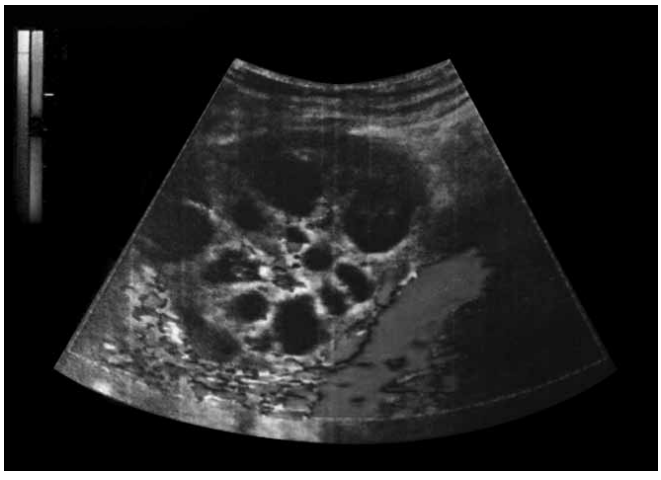

Fig. 2: Coronal section of CT showing large multiloculated cyst 13 × 8 × 6 cm in right adnexa extending up to lower abdomen, multiple septa seen. Three hypodense lesions in right lobe of liver seen

Intraoperatively (Fig. 3), the right ovary was enlarged 9 × 8 cm, torsion four times seen near right pedicle, specimen with areas of hemorrhage, congestion and stromal edema noted (Fig. 4). The left ovary and uterus were normal. Frozen section showed benign lesion with extensive areas of hemorrhage, congestion and stromal edema consistent with torsion, histopathology was reported as benign multiloculated cyst (Fig. 5).

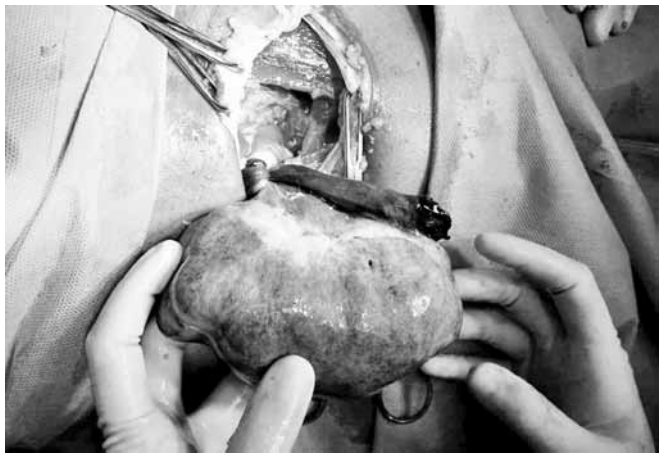

Fig. 3: Right ovary with torsion

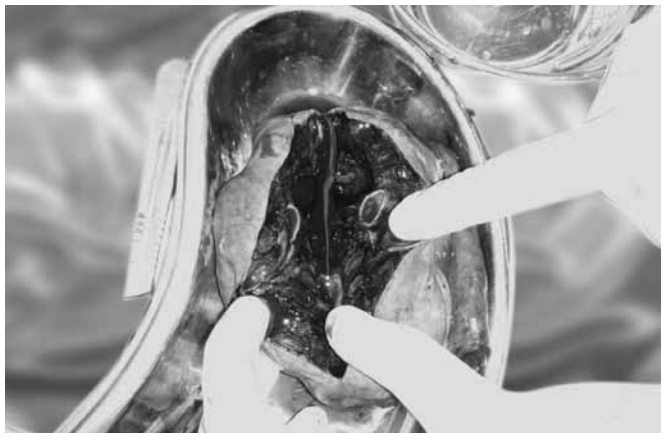

Fig. 4: Cut section of right ovary

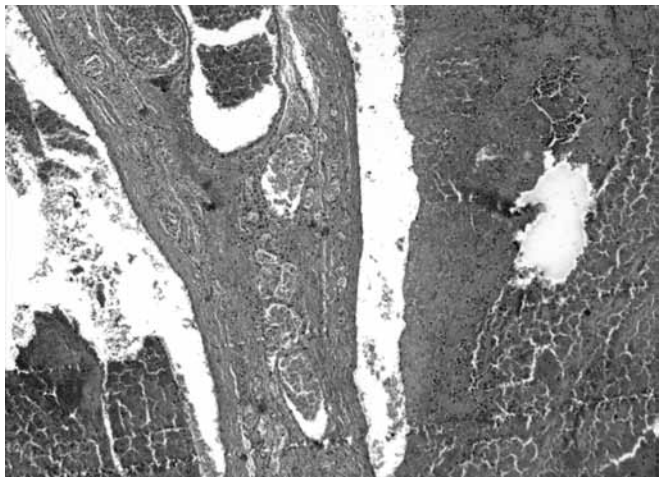

Fig. 5: Histopathology showing multiloculated cysts

Table 2: Case reports of hypothyroidism associated with multiple ovarian cysts

Author	*Yamashita*	*Taher BM*	*Bassam T*	*Kubota K*	*Present case*
Report year	2001	2004	2006	2008	2013
Age	19	22	19	21	21
C/O	Irregular cycles	Acute abd	Abd pain	Abd pain	Abd pain, distension
TSH	132.75	>100	4191	1840	>150
FSH	3.8	9.8	14	9.7	67.07
LH	1.6	12.6	1.1	<0.5	<0.07
Estradiol	1206	105.9	127.5	601	2230
Morpho of ovary	Multiple follicles 9 cm	Multiloculated cysts right 6 cm, left 5 cm	Multiple cysts right 4.5 cm, left 5 cm	Multiple cysts right 10 cm, left 4 cm	Multiple cyst 12 × 8 cm

DISCUSSION

The salient features of longstanding juvenile hypothyroidism include deficient linear growth, delayed puberty.[6] The syndrome consisting of primary hypothyroidism, precocious puberty and massive ovarian cysts was termed Van Wyk and Grumbach syndrome in 1960.[7] Several hypotheses about the mechanism of ovarian cyst formation associated with hypothyroidism have been proposed.

Firstly structural similarities between TSH, FSH, and their receptors may have some role. Extremely high levels of TSH can bind to FSHR and lead to activation of follicular cells (specificity spill over phenomenon).[8] A second explanation is changes in the gonadotropins. Patients with hypothyroidism may have relatively high FSH levels and low LH levels. Hypothyroidism is associated with slow GnRH pulse, slow GnRH pulse favor FSH production and secretion, whereas rapid frequency favor LH production.[9,10] Regression of both pituitary and ovarian cyst after administration of thyroid hormone has been reported.[11] Hypothyroidism is associated with functional ovarian hyperstimulation like syndrome. Spontaneous OHSS related with pregnancy has been described as depending on activating mutations of the FSH receptor (FSHR) gene, causing ovarian hyper-responsiveness to circulating FSH or even cross-responsiveness of FSHR to hormones having a structure similar to FSH, such as human chorionic gonadotropin (hCG) or TSH.[12,13] Similar cases have been reported from 2001 to 2008 as shown in Table 2 but this case has the highest FSH reported and estradiol levels that drastically declined with thyroxine therapy. This case report illustrates that health providers should have a high suspicion for hypothyroidism in patients with multiple ovarian cysts, these symptoms can be completely reversed with initiation of thyroid replacement, avoiding unnecessary surgery and anesthesia.

CONCLUSION

Multicystic ovaries, as a presenting feature of juvenile hypothyroidism, are rare. Appropriate diagnosis and thyroid replacement therapy is effective and prevents inadvertent surgery. The time course for regression of multicystic ovaries with thyroid replacement varies between 3 and 6 months in most case studies.[10,11] Serum TSH should be measured in every adolescent with menstrual irregularities.

REFERENCES

1. Hansen KA, Tho SP, Hanly M, Moretuzzo RW, McDonough PG. Massive ovarian enlargement in primary hypothyroidism. Fertil Steril 1997 Jan;67(1):169-171.
2. Lindsay AN, Voorhess ML, MacGillivray MH. Multicysticvaries in primary hypothyroidism. Obstet Gynaecol 1983 Apr;61(4):433-437.
3. Ghosh S, Kabir SN, Pakrashi A, Siddhartha C, et al. Subclinical hypothyroidism: a determinant of polycystic ovary syndrome. Horm Res 1993;39:61-66.
4. Rotmensch S, Scommegna A. Spontaneous ovarian hyperstimulation syndrome associated with hypothyroidism. Am J Obstet Gynecol 1989 May;160(5 pt 1):1220-1226.
5. Anasti JN, Flack MP, Froehlich J, Nelson LM, Nisula BC. A potential novel mechanism for precocious puberty in juvenile hypothyroidism. J Clin Endocrinol Metab 1995 Jan;80(1): 276-279.
6. Oddie TH, Boy CM, Fisher DA, Hales IB, et al. Incidence of signs and symptoms in thyroid disease. Med J Aust 1972 Oct 28;2(18):981-986.
7. Rastogi A, Bhadada SK, Bhansal A. An unusual presentation of a usual disorder: Van Wyk-Grumbach syndrome. Indian J Endocrinol Metab. 2011 Jul;15(Suppl 2):S141–S143.
8. Yoshimura M, Hershman JM. Thyrotropic action of human chorionic gonadotropic. Thyroid 1995 Oct;5(5):425-434.
9. Mistry DS, Tsutsumi R, Fernandez M, Sharma S, Cardenas SA, Lawson MA, Webster NJ. Gonadotropin-releasing hormone pulse sensitivity of follicle-stimulating hormone-beta gene is mediated by differential expression of positive regulatory activator protein 1 factors and co-repressors SKIL and TGIF1. Mol Endocrinol 2011 Aug;25(8):1387-1403.

10. Ferris HA, Shupnik MA. Mechanisms for pulsatile regulation of the gonadotropin subunit genes by GNRH1. Biol Reprod 2006 Jun;74(6):993-998.
11. Yamashita Y, Kawamura T, Fujikawa R, Mochizuki H, Okubo M, Arita K. Regression of both pituitary and ovarian cysts after administration of thyroid hormone in a case of primary hypothyroidism. Int Med 2001 Aug;40(8):751-755.
12. Vasseur C, Rodien P, Beau I, Desroches A, Gerard C, de Poncheville L, Chaplot S, Savagner F, Croue A, Mathieu E, et al. A chorionic gonadotropin-sensitive mutation in the follicle-stimulating hormone receptor as a cause of familial gestational spontaneous ovarian hyperstimulation syndrome. N Engl J Med 2003;349(8):753-759.
13. Montanelli L, Delbaere A, Di Carlo C, Nappi C, Smits G, Vassart G, Costagliola S. A mutation in the follicle-stimulating hormone receptor as a cause of familial spontaneous ovarian hyperstimulation syndrome. J Clin Endocrinol Metab 2004;89(3):1255-1258.