POLYCYSTIC OVARIES WITH STROMAL HYPERTHECOSIS: A RARE PRESENTATION

Shaila Sheraffudeen¹, Sujamol Jacob², Sithara S³, Shilpa M⁴

¹Professor, Department of Obstetrics & Gynaecology, SATH, MCH, TVPM. ²Additional Professor, Department of Obstetrics & Gynaecology, SATH, MCH, TVPM. ³Assistant Surgeon, Family Planning, Department of Obstetrics & Gynaecology, SATH, MCH, TVPM. ⁴Junior Resident, Department of Obstetrics & Gynaecology, SATH, MCH, TVPM.

ABSTRACT

Presenting a 47-year-old peri-menopausal lady with hypomenorrhoea, temporal baldness, alopecia, hirsutism. The histopathology was polycystic ovaries with stromal hyperthecosis. Hughesdon described hyperthecosis as a severe form of PCOS. Hyperthecosis is rare in young women, with clinical features similar to PCOS. However, these women are usually more virilised.

KEYWORDS

PCOS – Polycystic Ovarian Syndrome, BMI – Body Mass Index.

HOW TO CITE THIS ARTICLE: Sheraffudeen S, Jacob S, Sithara S, et al. Polycystic ovaries with stromal hyperthecosis: A rare presentation. J. Evid. Based Med. Healthc. 2016; 3(36), 1801-1803. DOI: 10.18410/jebmh/2016/403

INTRODUCTION: Ovarian stromal hyperthecosis is an unusual cause of virilization in women. Ovarian stromal hyperthecosis (SH) is characterized by the presence of luteinized thecal cells within the ovarian stroma separate from the follicles and is usually accompanied by stromal hyperplasia. It is important to define the aetiology of increased levels of androgens in women because of the possibility of malignancy originating from the ovaries or adrenal gland. Ovarian stromal hyperplasia (OSH) and ovarian hyperthecosis (OHT) are both similar and nonneoplastic pathologies. Stromal hyperplasia is the nodular or diffuse proliferation of the ovarian stroma, whereas ovarian hyperthecosis is stromal proliferation accompanied by luteinised stromal cells. They both show an excess of androgen production and have a wide clinical range, including hirsutism, virilisation, abnormal menses, obesity, hypertension and insulin resistance. This androgenic process leads to an elevation of testosterone. Additionally, by peripheral aromatisation of androgenic hormones, an increase in oestrogenic hormones occurs. The following case is an example of Ovarian stromal hyperthecosis which was evaluated and operated in our hospital.

CASE REPORT: A 47-year-old lady apparently normal till 8 years back presented with alopecia (Fig. 1), male distribution of body hair, FGS score – 22, hypomenorrhoea, frontal and temporal baldness, (Fig. 2) deepening of voice progressively increasing over past 8 years. Menarche was at 12 yrs. with regular cycles of 3-4 days/28-30 days till date of admission. She conceived spontaneously 6 months after marriage and had 3 full term normal deliveries with an uneventful antenatal and postnatal period. There was no treatment for infertility. She was diagnosed as hypertensive since 2 yrs.

Financial or Other, Competing Interest: None. Submission 30-03-2016, Peer Review 19-04-2016, Acceptance 27-04-2016, Published 05-05-2016. Corresponding Author: Dr. Shaila Sheraffudeen, Professor, Department of Obstetrics & Gynaecology, SATH, Government Medical College, Trivandrum. E-mail: shailas58@gmail.com DOI: 10.18410/jebmh/2016/403 There is no other medical illness. She was prescribed oral contraceptive pill containing cyproterone acetate which she took only for 6 months. As there was no improvement she came requesting for hysterectomy.

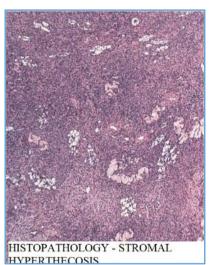


Fig. 1



Fig. 2

On Examination: BMI-29.5, there was frontal and temporal baldness, alopecia, male pattern body hair, chest and abdominal hair, clitoromegaly (Fig. 3), deepening of voice. Investigations: Blood investigations -Table 1, CT Abdomen - Bilaterally enlarged ovaries with follicles, few cystic changes.



Fig. 3

Intra Operative Findings: Bilaterally enlarged ovaries 5 x 3 cm (Fig. 4), solid tumour with cystic areas (Fig. 5).



Fig. 4



Fig. 5

Histopathological Report: Polycystic ovaries with stromal hyperthecosis (Fig. 6). Postoperatively, there is an improvement in alopecia and voice has normalised.

DISCUSSION: The "polycystic ovary syndrome" constitutes a functional and hormonal disorder. It is usually caused by several alterations in the functioning of the intricate mechanisms of the hypothalamus-pituitary-ovaries axis and sometimes including disorders in other areas of the endocrine system. The frequent clinical manifestations of the "polycystic ovaries syndrome" are: 1) Oligomenorrhoea and/or episodes of amenorrhea. 2) Hypertrichosis or hirsutism, frequently associated with acne. 3) Chronic anovulation and infertility. Some patients exhibit a tendency to weight gain or even obesity. The menstrual disorders may also include hypermenorrhoea and/or menorrhagia. Presently, some disorders in the insulin metabolism are being found in many women with "polycystic ovaries syndrome".⁽¹⁾ This fact is making many researchers attribute great importance to this "insulin-resistance" on the genesis of the syndrome, and they argue that this metabolic disorder can increase the production of androgens by the ovaries.

The fact is, both the ovaries and the hypothalamicpituitary function are deeply altered, creating a vicious circle. Besides the functional disturbance, the ovaries also exhibit considerable histologic and morphologic alterations, characterised by the hyperthecosis (hyperplasia of the ovarian stroma) (Fig. 7) and the bilateral enlargement of these organs. As it was also observed, an excessive production of androgens by the adrenal glands (hyperandrogenic adrenal hyperplasia) may also be responsible for several cases of "polycystic ovaries syndrome", and sometimes both conditions may be associated.



Fig. 6



Fig. 7

In 1982, Hughesdon suggested that hyperthecosis is a severe form of PCOS.⁽²⁾ Hyperthecosis is rarely seen in young women and is generally encountered in menopausal women.⁽³⁾ The clinical features of hyperthecosis are similar to PCOS. However, women with hyperthecosis have more hirsutism and are likely to be virilised.⁽⁴⁾ Most women with ovarian hyperthecosis are obese and have a long-standing history of hirsutism. The hirsutism is severe, and most of the women shave daily. Many also have clitoral enlargement, temporal balding, deepening of the voice and a male habitus. Most have amenorrhea, and the remainder have irregular and anovulatory cycles. Some have acanthosis nigricans, suggesting severe insulin resistance.

A familial occurrence of hyperthecosis has been reported, with the mode of inheritance being consistent with an autosomal dominant pattern. Women with hyperthecosis have severe insulin resistance and do not ovulate with clomiphene treatment. Ovarian stromal hyperplasia and hyperthecosis are commonly seen in postmenopausal women and involve both ovaries.⁽³⁾ In severe hyperthecosis, ultrasonography shows bilateral expansion in the ovarian stroma; ovaries appear more solid, and a few cysts may also be seen.⁽³⁾

CONCLUSION: Potential areas of further research activity include the analysis of predisposing conditions that increase the risk of PCOS, particularly genetic background and environmental factors, such as endocrine disruptors and diet.⁽⁵⁾

Intra-ovarian regulation of follicle development and mechanisms of follicle arrest and the impact of metabolic abnormalities on these processes, as well as molecular mechanisms by which insulin excess regulates androgen secretion and metabolism and disrupts follicle development⁽⁵⁾ are other potential issues for investigation.⁽⁶⁾

PCOS status may lead to many longterm consequences in women, specifically the development of type 2 diabetes. Identifying susceptible individuals would help to individualise therapeutic and, possibly, preventive strategies.

Serum testosterone	2.40	15-70 ng/dL (0.52-2.4 nmol/L)
Serum estradiol	23.2	Mid-follicular phase: 27-123 pg/mL Periovulatory: 96-436 pg/mL Mid-luteal phase: 49-294 pg/mL
Luteinizing Hormone	3.0	1-20 IU/L.
Follicle Stimulating Hormone	8.0	3-20 mIU/mL
DHEAS	96	35-430 ug/dL
17 Hydroxy Progesterone	2.0	20-100 ng/dL prior to ovulation, 100-500 ng/dL during the luteal_phase.
Thyroid Stimulating Hormone	0.85	0.4-4.5 μIU/mL
Serum Prolactin	12	13 ug/L
Table 1		

REFERENCES

- 1. Nelson Soucasaux. Polycystic ovary Syndrome. 2002.
- 2. Hughesdon PE. Morphology and morphogenesis of the Stein-Leventhal ovary and of so-called hyperthecosis. Obstet Gynecol Surv 1982;37(2):59-77.
- Sinan Beksac, İlker Selçuk, Gökhan Boyraz, et al. Two patients with marginal symptoms showing hyperthecosis at the edge of malignancy: presentation of two cases. Turkish-German Gynecol Assoc 2013;14(3):182-185.
- Barth JH, Jenkins M, Belchetz PE. Ovarian hyperthecosis, diabetes and hirsutism in postmenopausal women. Clin Endocrinol (Oxf) 1997;46(2):123-128.
- 5. Franks S, Stark J, Hardy K. Follicle dynamics and anovulation in polycystic ovary syndrome. Human Reproduction Update 2008;14(4):367-378.
- Diamanti-Kandarakis E, Bourguignon JP, Giudice LC, et al. Endocrine-disrupting chemicals: an endocrine society scientific statement. Endocrine Reviews 2009;30(4):293-342.