

EP_A138**XANTHOMATOUS HYPOPHYSITIS PRESENTING WITH PROGRESSIVE HYPOPITUITARISM AND PITUITARY APOPLEXY**

<https://doi.org/10.15605/jafes.039.S1.149>

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INTRODUCTION/BACKGROUND

Primary hypophysitis is a rare condition characterised by isolated inflammation of the pituitary gland and infundibulum. Among the histopathological subtypes, xanthomatous hypophysitis is rare.

CASE

We describe a 28-year-old female with xanthomatous hypophysitis (XH), its clinical course over 4 years, as well as the transsphenoidal surgery outcome.

She first presented 4 years ago with intermittent headaches and amenorrhea for four months. Investigations showed that she had central hypogonadism (FSH 0.9 IU/L, LH <0.1 IU/L, estradiol 77 pmol/L) and hyperprolactinemia (prolactin 2195 m IU/L). She continued to have central hypogonadism despite prolactin normalisation and was subsequently started on an oral contraceptive pill. A pituitary MRI showed a normal pituitary gland and no stalk lesion. One year later, she developed arginine vasopressin (AVP) deficiency, leading to desmopressin treatment. She was diagnosed with autoimmune hypophysitis, was started on prednisolone and 50 mg of azathioprine daily. However, she developed transaminitis after two months of treatment and it was withheld.

Two and a half years later, she developed central hypothyroidism and central adrenal insufficiency and was started on hydrocortisone and levothyroxine replacement. She was admitted to the ward two months later for pituitary apoplexy, and a repeat MRI showed an enlarging sellar cystic mass with mass effect and increasing T1-weighted hyperintensity within. The Humphrey visual field examination was normal. She underwent transsphenoidal hypophysectomy which revealed a sellar mass with a thickened capsule containing cheesy material. Histopathological findings were consistent with xanthomatous hypophysitis. Three months after surgery, she still had panhypopituitarism, and a repeat MRI revealed a pituitary gland within the sella with no evidence of recurrent disease.

CONCLUSION

Surgery is the treatment of choice for patients with XH who present with pituitary apoplexy. Close follow-up and monitoring of hormonal recovery and disease remission are essential to observe the clinical progression of this rare disease.

EP_A139**FERTILITY IN CONGENITAL ADRENAL HYPERPLASIA: A CASE REPORT**

<https://doi.org/10.15605/jafes.039.S1.150>

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INTRODUCTION/BACKGROUND

Congenital adrenal hyperplasia (CAH), an autosomal recessive disorder affecting cortisol biosynthesis enzymes, results in virilization in affected females. Fertility challenges are often faced by females with CAH. We present two women with CAH who achieved successful spontaneous pregnancy and their pregnancy outcomes.

CASE 1

Ms. NAS, a 28-year-old Malay female, was diagnosed with 21-hydroxylase deficiency (21-OHD) at birth (presentation: ambiguous genitalia) and underwent corrective surgery at one year of age. She attained menarche at the age of 12 before having secondary amenorrhea for one year at 13 years old. She had normal menstrual cycles while on hydrocortisone 10 mg BD. Pre-pregnancy investigations were as follows FSH 4.04 IU/L, LH 2.22 IU/L, testosterone 2.22 nmol/L, dehydroepiandrosterone Sulphate (DHEA-S) 1.040 umol/L (NR 2.68-9.23). She safely delivered her baby via spontaneous vaginal delivery.

CASE 2

Ms. NN, a 22-year-old female, was diagnosed with CAH at 1 month of life presenting with a salt-losing crisis. She developed precocious puberty at 9 years old due to poor compliance to treatment. She was on triptorelin (Decapeptyl) sc for 2 years until the age of 12. She successfully conceived at the age of 21. Pre-pregnancy, she was treated with T hydrocortisone 5 mg/5 mg/7.5 mg TDS and T fludrocortisone 0.1 mg OD. Her hormonal level preconception were as follows: testosterone <0.1 nmol/L, 17 hydroxyprogesterone (17 OHP) 71.9 nmol/L (elevated), DHEA-S 0.349 umol/L (suppressed). Her pregnancy was uneventful, and successfully delivered her baby surgically after having transverse lie at term.

The differences between the 2 cases (presentation of CAH, the need for genital reconstructive surgery, the nature of puberty) did not impair the fertility potential of these 2 patients during adulthood.

CONCLUSION

Management of CAH during pregnancy is important to ensure successful term delivery with no added pregnancy complications like gestational diabetes or hypertension.

EP_A140

VIRILISING OVARIAN TUMOUR: A TERTIARY CENTRE EXPERIENCE

<https://doi.org/10.15605/jafes.039.S1.151>

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INTRODUCTION/BACKGROUND

To determine the clinical features, a spectrum of imaging characteristics and histopathologic findings of virilizing tumours of the ovary.

CASE

A 69-year-old female presented with a 1-year duration of increasing hair growth over both arms and androgenic and male pattern facial hirsutism which needed facial hair trimming once a week.

She attained menarche and menopause at the age of 12 and 55 respectively. She was diagnosed with hypertension and Graves' disease at 30 years of age. She was clinically obese with a BMI of 30 kg/m², blood pressure 149/84 mmHg, deepening of voice and hirsutism score of 16. There was no clitoromegaly, no proximal myopathy, no cutaneous bruising or abdominal striae. The total testosterone level was >52.05 nmol/L (NR: 0.1-1.6) with normal ACTH, cortisol and DHEA-S. An abdominal CT scan showed a left ovarian mass measuring 7.3 x 7.4 x 5.4 cm with solid components and signs of peritoneal carcinomatosis; however, no calcification within the mass was noted. She underwent primary debulking total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAHBSO), omentectomy and pelvic node dissection.

Histopathological examination demonstrated steroid cell tumour of the ovary. Post-operatively, testosterone levels reduced to normal and showed improvement of alopecia and hirsutism on follow-up 6 months later.

CONCLUSION

In post-menopausal women the appearance of signs of virilization and high testosterone levels should be

investigated systematically for underlying malignancy and to determine if the high testosterone levels are of an adrenal or ovarian origin. Failure of recognition will lead to a poor prognosis. In this patient, hematological investigations ruled out an adrenal cause however radiological imaging identified an ovarian etiology.

EP_A141

WHAT TESTOSTERONE CONCENTRATIONS SHOULD WE EXPECT IN HEALTHY MEN?

<https://doi.org/10.15605/jafes.039.S1.152>

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INTRODUCTION/BACKGROUND

Older men generally have lower testosterone concentrations compared with younger to middle-aged men. In aging men, lower testosterone concentrations are associated with poorer health outcomes. Understanding the relationship between testosterone and men's health status has implications for defining appropriate testosterone reference ranges to apply in clinical practice.

METHODOLOGY

Studies defining reference ranges for testosterone in selected groups of healthy men and meta-analyses of prospective cohort studies which analysed associations of testosterone with key health outcomes in community-dwelling men were identified. These studies used mass spectrometry to assay testosterone concentrations.

RESULTS

In 124 men aged 21-35 years with normal reproductive function, 95% confidence limits for testosterone were 10.4-30.1 nmol/L. In 1185 non-obese men aged 19-39 years, the range was 9.2-31.8 nmol/L. In 394 healthy men aged 70-89 years, the range was 6.4-25.7 nmol/L. Individual participant data (IPD) meta-analyses of 21,074 men from nine studies showed lower testosterone concentrations in men older than 70 years (with higher luteinising hormone), and in those with higher BMI, or with diabetes or cancer. Extension of these IPD meta-analyses showed non-linear associations of testosterone with all-cause and cardiovascular mortality risk. Men with low testosterone concentrations (<7.4 nmol/L) had higher all-cause mortality.

CONCLUSION

Reference ranges for testosterone in healthy younger to middle-aged men are higher than for healthy older men.