*Singapore Journal of Obstetrics and Gynaecology*
*March 2004; 35(1): 53-57*

# CASE REPORT

# A RARE CASE OF VIRILIZATION IN PREGNANCY WITH SPONTANEOUS RESOLUTION POSTPARTUM

P Kannappan[1]
A R Azlina[2]

## CASE REPORT

A thirty-five year old Indian lady had presented in her third pregnancy at 31 weeks of gestation with hirsuitism, hoarseness of voice, coarse and dry skin, as well as enlargement of the limbs. She complained of reduced effort tolerance and orthopnea since 16 weeks of gestation. She weighed 70kg with a height of 148cm (BMI=31.95) and was normotensive (128/69). There was hirsuitism, thickened skin, spade like hands, large nose, goitre, normal intradental recess, hoarseness, of voice with carpel tunnel syndrome over the left hand and acanthosis nigricans over the axilla with clitromegaly. Neurological / cardiovascular / respiratory system and funduscope examination was normal. On per abdomen examination the fundal height corresponded with the gestation.

In the following two weeks she was experiencing pain at the pelvic joint with dark pigmentation of the finger nails. At 36 weeks gestation she complained of gum hypertrophy with difficulty during eating and brushing. The pain at the pelvic joint had worsened. Her total weight gain in pregnancy was 8.5kg.

Her booking was at 13 weeks gestation and the first hospital visit was at 26 weeks gestation. She and her sister had a milder version of similar symptoms in their prior pregnancies. She had an elective caesarean section for suspected big baby in 1993 and delivered a baby girl weighing 3.6kg. This was followed by vaginal delivery of a baby boy weighing 2kg in 1996. Both chlidren are well.

She underwent a range of investigations. The abnormal results were repeated in the postpartum period with CT imaging of the pituitary fossa and adernal gland which was normal. The results are as follows;

[1]*Department of Obstetrics and Gynaecology*
*Hospital Muar,*
*Malaysia*

[2]*Department of Pathology,*
*Hospital Muar,*
*Malaysia*

*Correspondence:*
*Dr P. Kannappan*
*Department of Obstetrics & Gynaecology,*
*Hospital Muar,*
*84000 Muar,*
*Johor,*
*Malaysia*

CASE REPORT

# A RARE CASE OF VIRILIZATION IN PREGNANCY WITH SPONTANEOUS RESOLUTION POSTPARTUM

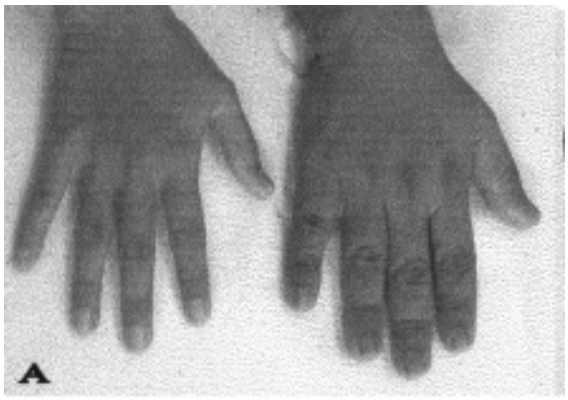



Fig.1. Comparison between patients right hand and a female ward staff's right hand.

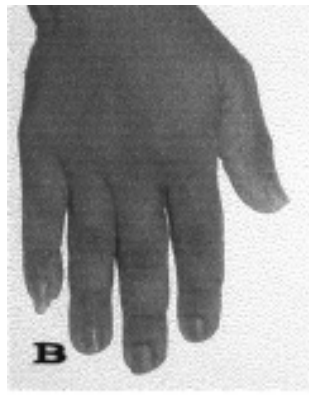



Fig.2. Patient's right hand in the postnatal period.

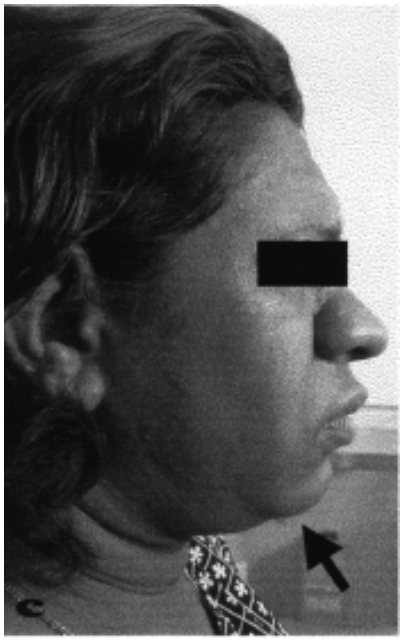



Fig.3. Lateral view in the antenatal period showing coarse facial features with hirsutism and skin thickening

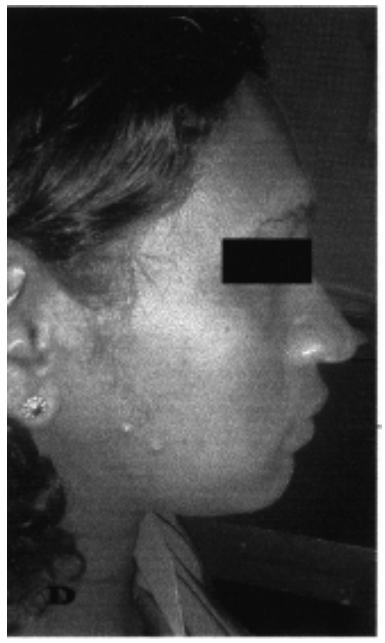
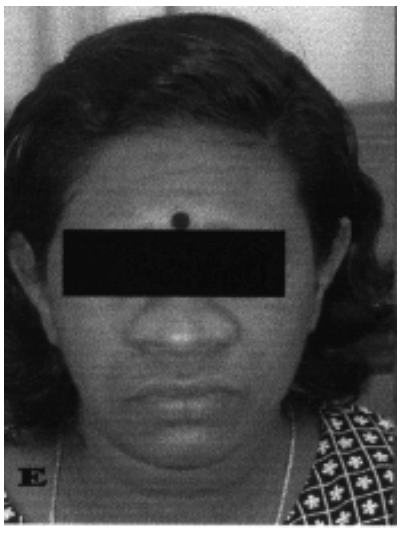



Fig.4. In the postnatal period



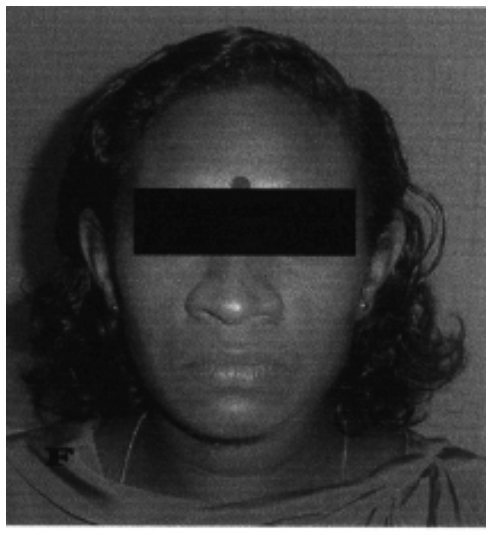




Fig.5. Facial features in pregnancy

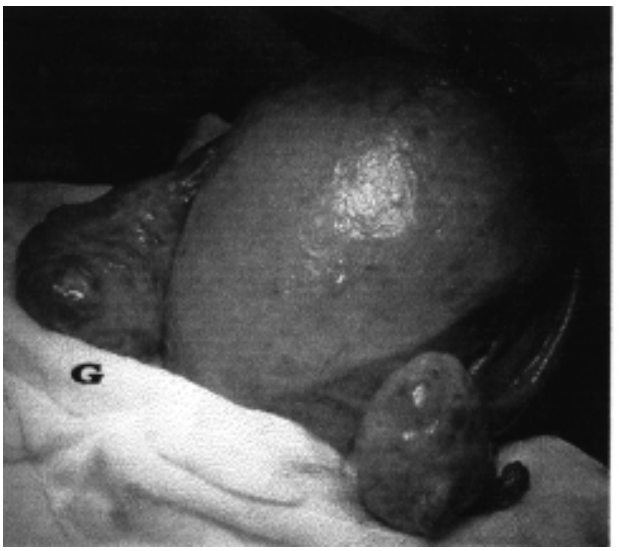


Fig.7. Showing the bilaterally enlarged polycystic ovaries

| During pregnancy | Postpartum |
|---|---|
| Blood count & Renal profile - normal | |
| Impaired MGTT(5.4/10.7mmol/L) with normal blood sugar profile. | Normal MGTT *(Modified Glucose Tolerance Test)* |
| Serum testosterone: 15.5nmol/L (elevated) | Serum testosterone: 1.1nmo1/L (normal) |
| FSH / LH – 7.75/5.17mlU/L (normal) | FSH / LH – 4.7/2.0mlU/L (normal) |
| TSH: 1.4miu/l (normal); Free T4: 0.74miu/l (low) | TSH: 0.34miu/1 (low); Free T4: 21.4pmo1/1 (normal) |
| Serum cortisol / growth hormone / DHEAS – normal | Serum cortisol / growth hormone - normal |
| Serum prolactin: 718.9mlU/L (slightly elevated) | Serum prolactin: 305mlU/L (normal) |
| CA125/CEA – normal | |
| Alpha Feto-protein: 377ng/ml (elevated) | Alpha Feto-protein: 4.0ng/ml (normal) |
| ECHO was normal (ejection fraction of 73%) | |
| Urine analysis - normal | Total cholesterol: 5.1mmo1/L(normal) |

She underwent an elective caesarean section with bilateral ovarian wedge biopsy and tubal ligation at 37 weeks gestation. The postoperative recovery was uneventful. She had a healthy baby boy weighing 3.4kg. Her symptoms subsided after delivery and so did the clinic features of virilization except for the voice change and clitromegaly. The histopathological report revealed possibility of hyperplastic lesion of the steroid cells such as leydig cell hyperplasia or hyperplastic of luteinized cells with theca luteal cysts. The microscopic findings were not suggestive of pregnancy luteoma.

## DISCUSSION

This lady had presented with signs and symptoms of virilisation with features of acromegaly, which had worsened in third trimester. She is a multiparous lady and had milder version of similar problem in the previous pregnancies. Intraoperatively there was bilateral polycystic enlargement of both ovaries. The serum testosterone level returned to normal postpartum. Mild degrees of hirsutism may appear around the 20th week of gestation but always completely disappear after delivery[1]. However features of virilization and acromegaly in pregnancy are rare. The prevalence of acromegaly in pregnancy is around 5 cases per 100,000 population[1] while among the cause for virilization is pregnancy luteoma of which only 100 cases have been described[2]. Polycystic ovarian disease is the most common cause of hirsutism in pregnancy[1] while there are only a few reported cases of it causing maternal virilization[3]. True androgen-secreting tumours are extremely rare in pregnancy[4,5]. Such diagnosis could be ruled out if ovarian venous catheterization showed elevated androgen level in both ovaries[3].

In this patient normal growth hormone levels followed by the imaging of the pituitary had ruled out the possibility of acromegaly. Adrenal causes are unlikely due to the normal DHEAS and serum cortisol. The clinical features were very suggestive of pregnancy luteoma where in 25% of cases virilization worsen during the latter half of pregnancy[2]. This is the result of elevated testosterone level with reduced testosterone binding proteins and it decreases rapidly after delivery. Among the differential diagnosis are granulosa tumours, thecomas and streoid cell tumours, which may occur during pregnancy, however such tumours are usually unilateral and solitary in nature. Tumours more frequently bilateral would be pregnancy luteoma, hilar leydig cell hyperplasia and maybe hyperplasia of luteinized cells. The latter have been associated with virilization in younger women, sometimes during pregnancy. In some cases it has been associated with elevated serum testosterone[2].

The infants are usually spared as in most situations the placenta aromatizes androgens to estrogens thus virilization of the female infants is unusual[1].

In most circumstances such lesion is benign, self-limiting and no further treatment is required.

## REFERENCES:

1. *Garner P.R. Pituitary and adrenal disorders; Turner M.L.C. The skin in pregnancy. In: Burro GN, Duffy TP, editors. Medical complications during pregnancy. Philadelphia: Saunders, 1999: 166.*

2. *Clement PB. Nonneoplastic lesion of the ovary. In Kurman RJ, editor. Blaustein's Pathology of the female genital tract. New York: Springer-Verlag: 1994:621-626.*

3. *Ben-Chetrit A. and Greeblatt E.M. Recurrent maternal virilization during pregnancy associated with polycystic ovarian syndrome: a case report and review of the literature. Human Reproduction 1995;10:3057-3060.*

4. *McClamrock H.D., Adashi E.Y. Gestational hyperandrogenism. Fertility and Sterility 1992;57:257.*

5. *Manganiello P.D., Adams L.V., Harris R.D., Ornvold K. Virilization during pregnancy with spontaneous resolution postpartum: a case report and review of the English literature. Obstet Gynecol Surv 1995 May: 50(5):404-10.*