

# TAMOXIFEN-INDUCED HIRSUTISM: AN UNUSUAL SIDE EFFECT IN A 5-YEAR OLD GIRL WITH McCUNE- ALBRIGHT SYNDROME

Heves Kırmızıbekmez, Gül Yeşiltepe Mutlu, Fatma Dursun, Şükriye Pınar İşgüven

Zeynep Kamil Obstetrics and Pediatrics Education and Research Hospital, Istanbul, Turkey

## Introduction

McCune-Albright Syndrome is a rare disorder defined as the triad of peripheral precocious puberty, café-au-lait skin pigmentation and fibrous dysplasia of bone, caused by mutation of the gene GNAS1, resulting in autonomous endocrine hormone excess.

Tamoxifen, a selective estrogen receptor modulator, has been used to treat the consequences of premature puberty such as vaginal bleeding and rapid bone maturation.

## Case report

❖ A 5-year and 3-month old girl was diagnosed with McCune-Albright Syndrome by the time she was 8 months old.

❖ She was admitted for further follow up.

❖ She had peripheral precocious puberty, café-au-lait skin pigmentation, fibrous dysplasia and hyperthyroidism

❖ She was receiving Methimazole and Cyproterone acetate.

❖ There was no complaint of vaginal bleeding or progression in breast tissue, but bone maturation was exceedingly accelerated.



❖ Rearranging the treatment with a selective estrogen receptor modulator, rather than Cyproterone, was decided. Tamoxifen was prescribed with a dose of 5 mg, twice a day.

❖ Approximately two months later, the patient came in with abnormal hair growth on the skin of her abdomen and back.

❖ Serum androgen levels were in normal ranges for age, ultrasound imaging for any tumoral lesion was negative and there was no evidence of exposure to any other substance. The pattern of terminal hair following the administration of tamoxifen and the exclusion of other possible causes suggested a possible side-effect to the drug.

## Table: Physical Examination and Laboratory Findings

Weight:	23 kg (1.61 SDS)
Height:	119 cm (2.11 SDS)
Bone Age:	11 years
Growth Velocity:	8 cm/year
Thyroid:	Stage-1 goiter
Skin:	Café-au-lait spots, with irregular borders, demonstrated a "respect of the midline".
Puberty:	Breast stage-5 in appearance, but glandular tissue was regressed
Distribution of Terminal hair:	Upper and lower parts of linea alba, cervical, lumbar and sacral regions of the back, femoral regions (Ferriman-Galway Score: 14)
Pelvic USG:	Right ovary: 1.8 ml, Left ovary: 8.9 ml including multiple cysts
Surrenal USG	Normal
FSH:	0.13 mIU/ml
LH:	<0.05 mIU/ml
Estradiol:	396 pg/ml
Total testosterone:	8.7 ng/dl
17-OH Progesterone:	1.1 ng/dl
1,4 Androstenedione:	0.5 ng/ml
DHEA-S:	4.6 µg/ml
Cortisole:	29 µg/dl

## Discussion

Unexpected onset of hirsutism and the exclusion of other possible causes suggested a possible side-effect to the drug. This is the first pediatric case of hirsutism due to Tamoxifen. Recently a 77 years old patient with breast carcinoma was reported to have hirsutism 8 weeks after the initiation of tamoxifen. This case was reported as "Tamoxifen-induced hirsutism" to the manufacturer(1). According to a medical analysis website, 8223 people reported having side effects while taking Tamoxifen citrate. Among them five patients (0.06%) had hirsutism (2).

- References:
1. Al-Niaimi F and Lyon C. Tamoxifen-induced hirsutism. J Drugs Dermatol. 2011;10:799-801
  2. Nolvadex Side Effects Center. Review: could Tamoxifen Citrate cause Hirsutism?. Feb, 26, 2014 (eHealthMe)



Figure: Abnormal hair growth on the skin of her abdomen, back and femoral regions.