

Case Report:

An unusual case of incomplete isosexual precocious puberty in a young girl with juvenile hypothyroidism

Pranab Kumar Sahana, Ashish Sudhakar Deshmukh, Nilanjan Sengupta, Chanchal Das, Ranen Dasgupta
Department of Endocrinology, Nilratan Sircar Medical College, Kolkata

ABSTRACT

Long-standing untreated primary hypothyroidism is an uncommon cause of praecocious puberty. The hypothyroidism, incomplete sexual maturation, galactorrhoea, and pituitary enlargement are reversed or corrected by levothyroxine therapy within a few months. We report a young girl with juvenile hypothyroidism who presented with early onset of menarche with failure of growth who was treated successfully with levothyroxine with resolution of early onset pubertal changes.

Key words: *Juvenile hypothyroidism, Isosexual precocious puberty, Multicystic ovaries*

Sahana PK, Deshmukh AS, Sengupta N, Das C, Dasgupta R. An unusual case of incomplete isosexual precocious puberty in a young girl with juvenile hypothyroidism. *J Clin Sci Res* 2014;3:188-90.

INTRODUCTION

Long-standing untreated primary hypothyroidism, usually a consequence of Hashimoto's thyroiditis, is an uncommon cause of incomplete isosexual precocious puberty. Juvenile hypothyroidism is a common disorder which usually presents with short stature and delayed puberty. Rarely sexual precocity can occur due to severe hypothyroid in young children. In girls precocity manifests as breast enlargement, uterine bleeding and multicystic ovaries. This condition is very important to recognize as it is completely treatable with levothyroxine. We present the unusual case of an incomplete isosexual precocious puberty in a seven-year-old girl with juvenile hypothyroidism who presented with early onset of menarche with failure of growth.

CASE REPORT

A seven-year-old girl presented with complaints of two episodes of vaginal bleeding in previous three months. Each episode lasted for two weeks. Bleeding was mild and subsided with

haemostatic drugs. Subsequently she was referred to us for evaluation. Parents also noticed that their child was not gaining height for last two years and she was shorter than her peers. She was born at term following a normal vaginal delivery. Her developmental milestones in early childhood were normal. There was no history of headache, vomiting, visual disturbance or galactorrhoea. She did not give a history of convulsion, meningitis, encephalitis, head trauma or use of any hormonal preparations. Her scholastic performance was average. There was no family history of precocious puberty or thyroid disease in the family.

On physical examination, her face was puffy. She had mild pallor and dry skin. Her height was 104 cm (< 3rd percentile) and weight was 19 kg (25-50th percentile). Her midparental sex adjusted height was 155.5 cm. Her breast and external genitalia were prepubertal. Pubic and axillary hairs were absent. Thyroid gland was not enlarged. Abdominal examination did not reveal any mass.

Received: 05 December, 2013.

Corresponding author: Dr. S. Pranab Kumar, Associate Professor, Department of Endocrinology, Nilratan Sircar Medical College, Kolkata, India. e-mail: pranabsahana@gmail.com



Online access

http://svimstpt.ap.nic.in/jcsr/jul-sep14_files/1cr314.pdf
DOI: <http://dx.doi.org/10.15380/2277-5706.JCSR.13.072>



Figure 1: Pelvic ultrasonography showing ovarian cyst with septae (arrow)

Laboratory investigations revealed normocytic hypochromic anaemia. Serum free T4 was 0.35 ng/dL (normal 0.89-1.76 ng/dL), thyroid stimulating hormone (TSH) was more than 750 mIU/mL (normal 0.5-4.5 mIU/mL). Serum prolactin was 66.9 ng/mL (1-20 ng/dL), follicle stimulating hormone (FSH) was 11.2 mIU/mL (normal 1.6-9.6 mIU/mL), luteinizing hormone (LH) was 0.15 mIU/mL (normal 1-11 mIU/mL), estradiol was 86 pg/mL (normal 20-60 pg/mL). Serum thyroid peroxidase (anti TPO) antibody levels were 256 IU/mL (normal < 35 IU/mL). Serum human chorionic gonadotrophin (HCG), alpha-foetoprotein (AFP) and carcinogenic antigen (CA-125) were normal. Pelvic ultrasonography revealed normal adult type uterus (6.3 × 2.9 × 2.1 cm) with a normal echotexture and endometrial thickness. Right and left ovaries showed cysts measuring of 3.0 × 1.8 cm and 5.4 × 5.2 cm respectively with multiple septae (Figure 1). Radiograph of wrist showed bone age of approximately five-and-half years which was delayed with reference to Greulich and Pyle method of assessment of bone age¹ (Figure 2). Radiograph of skull showed enlarged sella turcica.

Oral levothyroxine replacement was started. During follow-up at 6 months, she was well without any further vaginal bleeding. There was regression of size uterus and complete



Figure 2: Radiograph of left wrist for bone age assessment suggestive of bone age of about five-and-half years

disappearance of ovarian cysts on ultrasonography.

DISCUSSION

Isosexual precocious puberty is a rare manifestation of long-standing untreated juvenile hypothyroidism which was first described by Van Wyk-Grumbach in 1960.² In this disorder, girls present with breast enlargement and premature menarche. Unlike other precocious puberty, height acceleration does not occur in this condition and bone age gets retarded. Another case of case of isosexual precocity due to juvenile hypothyroidism has also been reported.³

Various mechanisms have been proposed to explain precocious puberty caused by hypothyroidism. It has been proposed that human TSH and human FSH act through the same receptor, namely the human FSH-receptor. This provides a potential novel mechanism for the precocious puberty of juvenile hypothyroidism.⁴ Other mechanism is overproduction of prolactin caused by overlap with the action of thyrotropin releasing hormone on the pituitary, resulting in either an increased level of gonadotropin-releasing hormone (leading to increased production of FSH and LH) or an increased level of oestrogen by upregulation of FSH receptors.⁵

Girls with acquired primary hypothyroidism may also present with abdominopelvic masses due to large ovarian cysts caused by hyperstimulation.⁶ Our patient had isolated menarche without breast enlargement and bilateral ovarian cysts. She also had short-stature and retarded bone age. During follow-up she had no more vaginal bleeding and ovarian cysts disappeared.

It is important to recognize this syndrome as symptoms regress with thyroid hormone replacement and patients spontaneously enter true puberty at an appropriate time.⁷ It is important to remember that hypothyroidism generally presents with delayed puberty but rarely it can present with precocious puberty which is completely reversible on treatment.

REFERENCES

1. Greulich WW, Pyle SI. Radiographic atlas of skeletal development of the hand and wrist. Second edition. California: Stanford University press;1959.
2. Van Wyk JJ, Grumbach MM. Syndrome of precocious menstruation and galactorrhoea in juvenile hypothyroidism: an example of hormonal overlap in pituitary feedback. *Pediatr* 1960;57:416-35.
3. Bhansali A, Kashyap A, Lodha S, Kotwal N, Ganapathi B, Mittal BR, et al. Isosexual precocity: uncommon presentation of a common disorder. *Postgrad Med J* 2000;76:177-8.
4. Anasti JN, Flack MR, Froehlich J, Nelson LM, Nisula BC. A potential novel mechanism for precocious puberty in juvenile hypothyroidism. *J Clin Endocrinol Metab* 1995;80:276-9.
5. Costin G, Kershner AK, Kogut MD, Turkington RW. Prolactin activity in juvenile hypothyroidism and precocious puberty. *Pediatrics* 1972;50:881-9.
6. Indumathi CK, Bantwal G, Patil M. Primary hypothyroidism with precocious puberty and bilateral cystic ovaries. *Indian J Pediatr* 2007;74:781-8.
7. Sharma Y, Bajpai A, Mittal S, Kabra M, Menon PS. Ovarian cysts in young girls with hypothyroidism: follow up and effect of treatment. *J Pediatr Endocrinol Metab* 2006;19:895-900.