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Case Report

Association between Michelin Tire Baby Syndrome and Congenital Panhypopituitarism in an Iranian Girl

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Abstract

Michelin tire baby syndrome is a rare syndrome, diagnosed clinically by multiple circumferential skin folds. Multiple noncutaneous anomalies have been described with this syndrome. We report a case of Michelin tire baby syndrome with congenital panhypopituitarism. To date, there is no report of association between these two disorders.

Keywords: Micheline Tire Baby syndrome, panhypopituitarism

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Introduction

Michelin tire baby syndrome (MTBS) is a rare genetic syndrome, first described in 1969 by Ross¹ and so named because of its resemblance to the “Michelin Man” logo of the French tire manufacturer.² It is characterized by generalized folding of excess skin² although it may be associated with additional phenotypic abnormalities. Diagnosis of this syndrome is mainly clinical and its exact pathogenesis remains unknown.^{2,3}

To date, there is no report of an association between MTBS and panhypopituitarism. Here, we describe a 7-month old Iranian girl presenting with this syndrome and panhypopituitarism.

Case Report

Our patient was a 7-month old girl, born from related parents (first degree parental consanguinity). She was normally delivered at term after an uncomplicated pregnancy, with a birth weight of 2.9 Kg, length of 50 cm and head circumference of 37 cm. When referred to our clinic, her weight was 5 Kg, her length was 52 cm and her head circumference was 42 cm. Multiple skin folds were noted on the upper and lower extremities. Skin thickness was normal and no hypertrichosis was seen (Figures 1 and 2). She had been previously diagnosed with panhypopituitarism and she was taking hydrocortisone (5mg/day), levothyroxine (0.025 mg/day) and growth hormone (Nordilet) 0.3 IU daily. We did not take any skin biopsy from our patient.

Discussion

The Michelin Tire baby syndrome is a clinical diagnosis, manifested by excessive folding of the skin.⁴ Various underlying abnormalities of mesenchymal tissues are diagnosed in this syndrome.⁵

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Both familial and sporadic cases of this syndrome have been reported. In familial cases, it has an autosomal dominant mode of transmission⁶ MTBS has been reported in otherwise healthy individuals and in association with various anomalies including thickened epiglottis,⁴ craniofacial anomalies, cleft palate, hypoplastic scrotum, inguinal and umbilical hernias⁷ congenital heart defect⁸ hemiplegia, microcephaly,⁹ and stellate scarring.¹⁰ Developmental delay, mental retardation and seizure have also been reported.^{1,2} The most common histologic features reported in affected skin of MTBS sufferers are nevus lipomatosus and smooth muscle hamartoma (with or without overlying hypertrichosis).^{2,4} Both adipose tissue and muscle originate from the mesoderm. Sato, et al., reported a case of MTBS patient, whose histology showed fragmented elastic fibers in addition to smooth muscle hamartoma.¹¹

Some MTBS cases have abnormal cytogenetic findings; including deletion of chromosome 11 and paracentric inversions of the long arm of chromosome 7.⁴ MTBS has a self-healing potential.²

Here, we report a case of MTBS with congenital panhypopituitarism (CPHP). Congenital hypopituitarism is a rare condition, characterized by multiple pituitary hormone deficiency including somatotroph, thyrotroph, lactotroph, corticotroph, or gonadotroph.¹² The signs and symptoms of CPHP are a combination of individual hormone abnormalities and may be nonspecific in early neonatal period with poor feeding, lethargy, apnea, jitteriness, hypoglycemia, temperature instability, poor weight gain, and prolonged neonatal jaundice.¹³ To date, there is no report of a relationship between CPHP and MTBS, although associations of developmental delay and growth retardation have been reported with MTBS syndrome without mention of the growth hormone level. Both MTBS and CPHP are rare disorders and further studies are needed to clarify whether this association is accidental or not. Based on our findings and previous reports of some phenotypic abnormalities associated with MTBS, such as hernias and thickened epiglottis, measurement of some pituitary hormones (growth hormone) and thyroid hormones may be useful in MTBS sufferers.

Conflict of interest: none



Figure 1. Prominent skin folds on the upper and lower extremities of the girl.



Figure 2. Closer view of skin folds of the upper extremity.

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