CASE REPORT

Cleft Palate Associated with Michelin Tire Baby Syndrome: A Rare Case Report

Sulakshana Pradeep[1], Sarakanuru Krishnappa Srinath[2], Padmapriya Surendranath[3]

Abstract

Michelin tire baby syndrome (MTBS) is one of the rare genodermatosis that is characterized by phenotypic abnormalities and multiple symmetric circumferential folding of excess skin. Diagnosis is done mainly through clinical examination. Skin fold gradually diminishes and fades away with age without any clinical intervention. Here, we are reporting a case of MTBS in a newborn.

Keywords: Cleft palate, Genodermatosis, Michelin tire baby syndrome.

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Introduction

The term Michelin tire baby (MTB) is adopted from the well-designed cartoon character of the Michelin Tire Company and it is characterized by the presence of multiple symmetric circumferential rings of folded skin described by Ross in 1969.[1] According to Classification in the Online Mendelian Inheritance in Man (OMIM), the phenotype MIM number for Michelin tire baby syndrome (MTBS) is 156,610 (skin creases, multiple benign ring-shaped, of limbs).[2] It is a rare syndrome and can be inherited as an autosomal dominant pattern, this syndrome is also known as "generalized folded skin". At birth, as the name implies, the skin is thrown into folds, but it is not lax and feels rather thick. With age, the skin folds spontaneously improve and in childhood, these are mainly confined to the extremities. It may be associated with various phenotypic abnormalities, and it may be a part of various syndromes.[3]

Case Description

An 11-day-old male child was referred to the Department of Pediatric and Preventive Dentistry from Vani Vilasa Hospital, Victoria Campus Bengaluru with a chief complaint of difficulty in feeding (Fig. 1). History suggested that child was delivered apparently vaginally with a birth weight of 2.9 kg, cried immediately after birth, born to second degree consanguineously married couple with the married life of 6 years. There was no positive family history. Mother was P1L1A0 (one living child), conceived after treatment for infertility with one 1st trimester abortion before this pregnancy. She had "Pregnancy induced hypertension" under control. Antenatal scans in the third trimester showed oligohydramnios (AFI = 4), with fetal MRI showing corpus callosum agenesis with bilateral severe lateral ventriculomegaly.

On physical examination, the length of the baby was 50 cm and head circumference was 33 cm, baby had dysmorphic facial features with brachycephaly, flat face, retrognathia, low set ears, thick helix, hypertelorism, periorbital fullness, blepharophimosis (underdeveloped eyelids) with short palpebral fissures, upslanting palpebral fissure, broad and depressed nasal bridge, microstomia, posterior cleft palate (Fig. 2), short neck with circumferential skin folds in the neck, long fingers, symmetrical circumferential skin folds in all the limbs (Fig. 3), abnormal genitalia-male pattern with scrotum anteriorly with unilateral undescended testis and penile shaft in the posterior part with hypospadias (opening of the urethra is not located at the tip of the penis) (Fig. 4). No systemic abnormalities were detected. On intraoral examination, cleft palate is noted involving hard and soft palate.

Investigations including cranial ultrasound, 2D echo, USG abdomen, and MRI brain were done. In cranial ultrasound-non communicating hydrocephalus-bilateral and lateral, 3rd and 4th

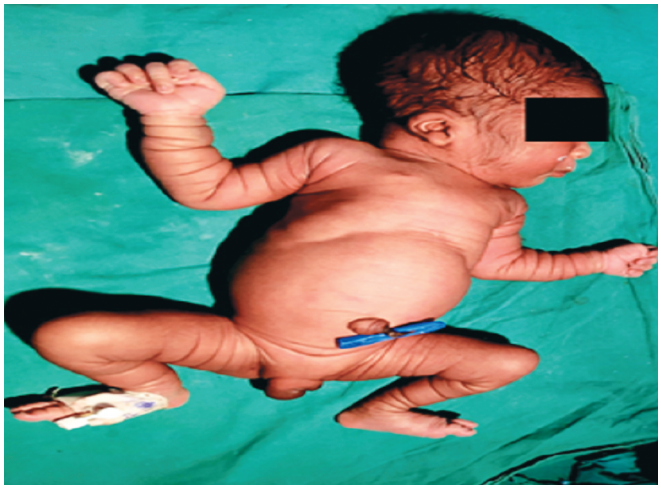

Fig. 1: Baby with Michelin tire baby syndrome

[1–3]Department of Pedodontics and Preventive Dentistry, Government Dental College and Research Institute, Bengaluru, Karnataka, India

Corresponding Author: Padmapriya Surendranath, Department of Pedodontics and Preventive Dentistry, Government Dental College and Research Institute, Bengaluru, Karnataka, India, Phone: +91 9497293318, e-mail: Padmapriya.vtm@gmail.com

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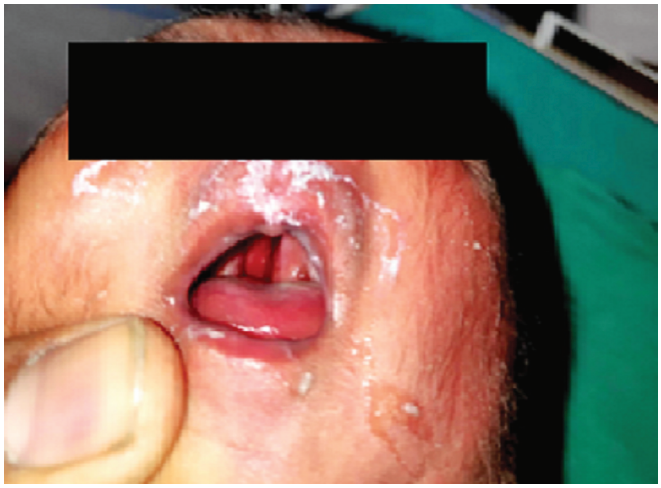

Fig. 2: Cleft palate

Fig. 3: Circumferential skin folds

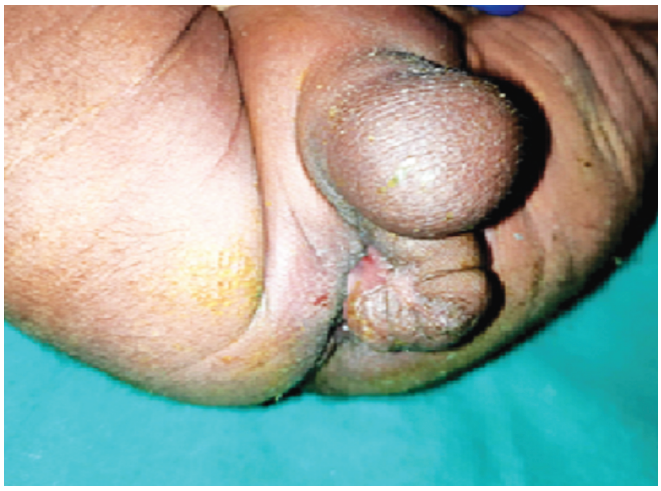

Fig. 4: Abnormal genitalia

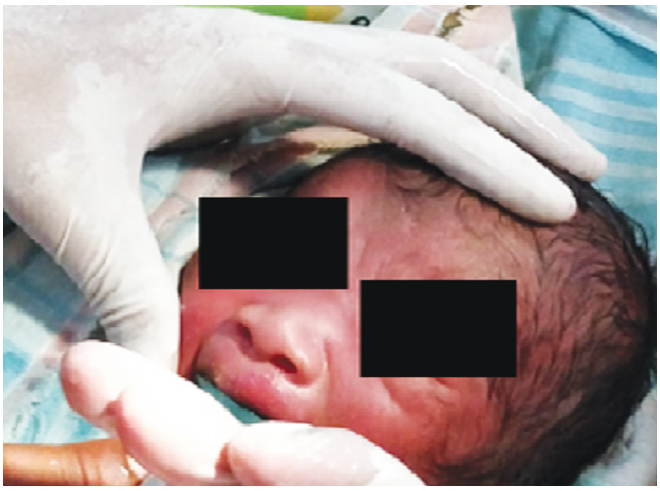

Fig. 5: Impression taking for obturator

Fig. 6: Obturator

ventricle dilatation was seen. Based on clinical appearance and investigations, a diagnosis of MTB syndrome was made in the Department of Pediatrics, Vani Vilasa Hospital, Bengaluru.

For 10 days, tube feeding was done and then the child was referred to our department for making an obturator. On the 11th day, impression of the cleft palate was taken with green stick impression material (Fig. 5). The cast was poured with dental stone and the obturator was prepared with acrylic material (Fig. 6). Appliance insertion was done on the 13th day. We demonstrated feeding with an appliance. Instructions were given regarding the cleaning of the obturator. Follow-up was done after 2 days. The baby was comfortable while feeding and there was no ulceration seen. Written consent was taken for both treatment and publication before the treatment was initiated.

Discussion

Michelin tire baby syndrome is characterized by the folding of excess skin that is generalized and may be associated with additional phenotypic abnormalities. The most common site of involvement is extremities, but it can also involve the trunk, palms, and soles. The pathogenesis is yet unclear. The condition may be familial. Cases of an autosomal dominant mode of inheritance in families have been reported.[4] Deletion of the short arm of chromosome 7 and paracentric inversion of the long arm of chromosome 11 and has been identified in association with MTBS.[5,6]

Michelin tire baby syndrome may be associated with various other congenital anomalies as well as other syndromes. Associated congenital anomalies are craniofacial anomalies, hypoplastic scrotum and hernias (inguinal and umbilical), left-sided hemihypertrophy, cleft palate, hemiplegia and microcephaly, smooth muscle hamartoma, joint hypermobility, epilepsy, stellate scarring, developmental delay, and psychomotor retardation.[5] Diagnosis of this syndrome is mainly through clinical examination, and the exact pathogenesis of this syndrome is unknown.[7]

It may be associated with other disorders such as Beare–Stevenson cutis gyrate syndrome associated with dermatomegaly localized to scalp, neck, face, and forehead, multiple congenital anomalies/mental retardation syndrome, undescended testis, hearing impairment, circumferential skin creases, and mental handicap syndrome.[8] A case was reported in which MTBS had a rare association with achondroplasia in which both of them were inherited as an autosomal dominant trait.[9] Michelin tire baby syndrome with derma sclerosis was also reported in the literature.[10]

The child was referred to genetic counseling at NIMHANS, Bengaluru to know the probable course of the syndrome and for options to deal with the recurrence of the syndrome. This is the first reported case of management of feeding difficulties associated with a cleft palate for a child with MTBS in a pediatric dental setup.

Conclusion

Michelin tire baby syndrome is one of the rare genodermatosis associated with cleft palate. Early diagnosis and management of these children are very crucial. A pediatric dentist plays an inevitable role in the multidisciplinary management of syndromic children.

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