

## Congenital Chikungunya with Centro-facial Pigmentation and Persistent Thrombocytopenia: A Case Report

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### Abstract

Hyperpigmentation over face in a neonate is rare and the differentials for the same are also rare. Congenital chikungunya, fungal and viral infections, drug rash are few differentials. Chikungunya Virus (CHIKV) infection manifesting in neonates is very rare. The prevalence of the entity was described only recently. We describe a neonate with hyperpigmentation on day 3 of life with stormy course thereafter. The distinguishing rash on face helped us in clinching the diagnosis of congenital chikungunya and fungal sepsis. Identification of this entity was based on characteristic skin rash and epidemiological background.

**Key Words:** Congenital chikungunya, Hyper pigmentation, Neonate.

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## Introduction

Chikungunya in neonates is a very rare entity and a diagnostic challenge. The clinical manifestations rarely resemble fulminant sepsis, making clinical decision and prognostication difficult. We report a case of mixed infection of congenital chikungunya and fungal sepsis with hyperpigmentation, the diagnosis of chikungunya was made on characteristic rash and epidemiological grounds (1).

## Case Report

A term appropriate for gestational age male baby was referred on day 14 of life, from Barshi, Maharashtra, Western India, with suspected sepsis and persistent thrombocytopenia.

Baby was born to a 26-year-old primi mother by normal vaginal delivery. Antenatally mother had fever 5 days prior to delivery. Immediate postnatal period was uneventful. On the 3rd day of life, parents noticed that the baby was warm, lethargic and had turned dark, hence was admitted in neonatal intensive care unit. He developed high-grade fever (103°F) on the same day. He was started on antibiotics and other supportive measures presuming sepsis. By the 5th day of life, fever had decreased in intensity. On the 6th day, baby was irritable. Platelet transfusions were given for 3 days in view of low platelet count. His thrombocytopenia and paradoxical cry was persistent, hence he was referred on the 14th day of life to our center.

At the time of admission, baby was irritable and had hyperpigmentation all over body but predominantly on face around nose (Fig.1). There was no

hepatosplenomegaly or other significant clinical features. Differentials kept were sepsis, congenital lupus, drug rash and addison's disease. Investigations showed the following: a markedly elevated C-reactive protein (111 mg/dl), hemoglobin of 12 g/dl, total leukocyte count 12700/mm<sup>3</sup>, differential count – lymphocytosis (78%), platelet count 70000/mm<sup>3</sup>, normal prothrombin and activated partial thromboplastin time, serum electrolytes, creatinine and liver function tests were normal. Blood culture grew *Candida Albicans*. Cerebrospinal Fluid (CSF) study was normal. Supportive treatment and injection Amphotericin B was given. Laboratory parameters also normalized over the next 2 weeks. Magnetic Resonance Imaging (MRI) done was normal. He did not have hypoglycemia or hypotension requiring inotropes and his hyperpigmentation was all over body, but predominantly on face around nose.

On detailed evaluation of history, antenatally, mother had high-grade fever and severe multiple joint pains 5 days before delivery. Mother continued to be symptomatic with severe arthralgia in the postnatal period as well. She did not have hyperpigmentation.

Similar clinical condition was reported to be present in the nearby areas and it was found that chikungunya was frequently diagnosed in that particular area. Mother was tested for chikungunya immunoglobulin M and immunoglobulin G, both of which were reported positive. However, the serology of the baby was reported negative. The diagnosis of congenital chikungunya was made based on the typical clinical manifestations in the

mother and baby and on the strong epidemiological profile. With supportive management, the baby improved, hyperpigmentation decreased. However, the irritability and paradoxical cry persisted until the discharge of the baby, most likely because of joint involvement. Baby was given paracetamol for the same. He was discharged after 2 weeks of intensive care stay and kept under follow-up.



**Fig.1:** Centropacial hyperpigmentation

## Discussion

Chikungunya fever (CHIKF) is an acute febrile illness caused by an arthropod-borne alphavirus, CHIKV. Available data suggest that CHIKV can be both endemic and epidemic (2). A maternal to child transmission of the disease has been described, although it is very uncommon. During the epidemic peak in Reunion Island, the attack rate was as high as 8.3% in pregnant women (3). In these instances, a higher risk for abortion in the first trimester has been reported compared to when the disease was acquired in the last trimester (4).

Neonatal infection could be associated with fever, poor feeding, tenderness,

unexplained apnea, distal edema, and various skin manifestations (5). Severe illnesses have been observed with associated encephalopathy, including pathologic MRI findings (brain swelling; cerebral hemorrhages) and possible evolution toward persistent disabilities.

In the acute stage, diagnosis is possible with Reverse Transcription Polymerase Chain Reaction (RT-PCR) or serology. Anti-CHIKV Immunoglobulin M (IgM) antibodies are detectable after an average of 2 days (1–12 days) by Enzyme-linked Immunosorbent Assay (ELISA) and remains positive for several weeks to 3 months (6). Immunoglobulin G (IgG) antibodies can be detected in the convalescent samples a few weeks later and persist for years (7). CHIKV infection seems to provide long-lasting protective immunity. The mother in this case was a confirmed case of chikungunya with typical clinical features and positive test results for IgM and IgG. She had severe arthralgia for a long duration after the pregnancy, requiring complete bed rest.

Although the serological test was negative in the baby, the diagnosis that was made in the mother along with the typical clinical features facilitated the diagnosis of chikungunya in the neonate. The paradoxical cry in the baby could be due to multiple joint inflammations. Baby had unexplained severe hyperpigmentation to start with. Differential diagnoses include congenital lupus, drug rash (eg. imipenem), and bacterial (*Listeria*, *S. epidermidis*), fungal (*Candida*) and viral (human herpes virus 6, enterovirus) infections. However Brownie nose suggested the possibility of chikungunya.

When we reviewed literature similar cases with hyperpigmentation have been reported in neonatal chikungunya, our case had both chikungunya and fungal sepsis. Typical Brownie nose in a neonate from endemic area with persistent thrombocytopenia, most likely differential diagnosis is chikungunya (5, 8, 9).

### **Conclusion**

Centro-facial hyperpigmentation is typical of congenital chikungunya and it has been described as brownie nose or chick sign. There are few differentials of Brownie nose – Congenital lupus, drug rash, bacterial, fungal and viral infections. To reach diagnosis, along with clinical presentation, it is important to know about epidemiological background.

### **Contributions**

Case report preparation and literature search by Dr. Kalane Shilpa. Manuscript editing and manuscript review by Dr. Rajhans Arti and Dr. Joshi Rajan

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