CASE REPORT

# Perinatal Chikungunya – A Report of 2 Cases

**Sajina Sathyan[a], Femitha Pournami[a], Anila V Panackal[a], Anand Nandakumar[a], Jyothi Prabhakar[a], Naveen Jain[a], PMC Nair[a]**

a. Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India

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**CORRESPONDING AUTHOR**

Dr Sajina Sathyan, MBBS, MD, DNB Neonatology Resident, Department of Neonatology, Kerala Institute of Medical Sciences Trivandrum, Kerala, India
Mobile: 7012696541
Email: sajinasathyan1988@gmail.com

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**AUTHOR INFORMATION**

1. Sajina Sathyan, MBBS, MD, DNB Neonatology Resident, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India.
2. Femitha Pournami, MBBS, MD, DM, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India
3. Anila V Panackal, MBBS, DCH, DNB, IAP Neonatology Fellowship, Registrar, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India
4. Anand Nandakumar, MBBS, MD, DNB, Consultant, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India.
5. Jyothi Prabhakar, MBBS, DCH, DNB, IAP Neonatology Fellowship, Consultant, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India.
6. Naveen Jain, MBBS, MD, DM, Senior Consultant, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India.
7. PMC Nair, MBBS, MD, DCH, DNB, DM, FIMSA, FRCP, Fellowship Neonatology, Department of Neonatology, Kerala Institute of Medical Sciences, Trivandrum, Kerala, India.

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## ABSTRACT

Chikungunya virus (CHIKV) infection manifesting in neonates is very rare. We describe 2 neonates with chikungunya one of whom presented with severe thrombocytopenia and features of multisystem involvement; diagnosis and management were difficult. Identification of this entity based on clinical background is challenging many times. The incidence of Chikungunya especially during the rainy season, family history and the typical hyperpigmentation on the nose and face (the CHIK sign) should be kept in the back of the mind for an early diagnosis and management.

## INTRODUCTION

Chikungunya in neonates is a comparatively rare entity. Clinical diagnosis may be challenging when clear epidemiological setting is lacking. Here we report 2 neonates with chikungunya, one of whom presented with multisystem involvement.

## CASE REPORT 1

This was a 32 weeker,1.8 kg girl baby born to 27 year old primi mother by emergency caesarean section in view of maternal fever and fetal decelerations. There was history of high grade fever in other family members 1 week prior. Mother developed pulmonary oedema on second postnatal day but subsequently improved.

In the immediate postnatal period, baby remained stable. Standard pre-term care given including continuous positive airway pressure, surfactant, donor human milk and antibiotic Amikacin for sepsis. By day 3, baby developed progressive abdominal distension, vomiting, melena and altered bloody aspirates. Baby remained intubated, was kept nil per oral and antibiotic was upgraded to Piptaz, This feed intolerance persisted till the end of third week of life. Features of capillary leak including abdominal distension, oedema over extremities, genital oedema were noted in the initial 3 weeks of life. There was persistent encephalopathy noted from third day to third week of life.

Investigations revealed platelet count of 13000 on third day of life which continued to be below 50000 till third week and baby was given 12 platelet transfusions in this period.

Hb was 9 on day 5, and packed cell transfusion given in view of the same. Initial CRP was negative but was 55 by day 7. Blood and CSF counts were negative, cultures were sterile. X ray abdomen was gasless. Ultrasound abdomen showed bowel wall oedema, minimal ascites and Superior mesenteric Artery (SMA) axis was normal. Blood PCR panel done by the end of first week of life showed low levels of Acinetobacter and Aspergillus. Antibiotics were upgraded to Meropenem and Amphotericin B. Viral film array panel and dengue NS1 were negative for mother.

By day 10, skin peeling of extremities and an eschar like rash were noted. Pus culture showed no growth. Between day 12-17, symptoms worsened and antibiotics were changed to Vancomycin, Colistin and Cefoperazone. Repeat blood PCR and other cultures showed no growth. Serum electrolytes/ ALT/ Creatinine were normal. Albumin infusion was given twice as albumin was 1.5 gm%.

From day 18 onwards baby started getting better. Respiratory supports were weaned; activity got better, oedema started reducing, skin lesions were healing, feeds could be increased and platelet counts started improving, following which antibiotics were stopped.

By day 21, the characteristic hyperpigmentation of nose, the "chik sign" was noted. The mother's Chikungunya IgM was tested which came positive. Baby was not tested as she received multiple transfusions.

Baby was discharged by 6 weeks and now baby is around 1 year of age. She is developmentally normal for her age.

## CASE REPORT 2

This was a term 38 weeker, 3 kg girl baby born to 26 year old primi mother by emergency caesarean section in view of fetal tachycardia. Mother had fever 1 day prior to delivery. There was history of fever and joint pain for father also. Immediate postnatal period was uneventful. On day 3 baby was shifted to intensive care unit in view of lethargy and hypoglycemia. She progressed to severe encephalopathy in next few hours along with recurrent apnoea for which she got intubated. Generalised erythematous rash was present which disappeared completely by next day.

Blood counts were normal. CRP done was negative. Blood and CSF cultures were sterile. Blood and CSF chikungunya PCR sent for baby which turned out positive. Chikungunya PCR for mother also was positive. Baby had loose stools which subsided by the end of first week. Baby could be extubated in the next 3 days and encephalopathy improved over 2-3 days thereafter. Baby was discharged by day 10. MRI brain showed symmetrical diffuse areas of diffusion restriction involving the subcortical and deep white matter of fronto-parieto temporal regions, corpus callosum internal and external capsule, anterior commissure, and postero-lateral thalamus. Baby is under follow up.

**Table 1. Skin Manifestations**

- Maculopapular rash
- Blotchy nasal erythema
- Freckle like pigmentation over center of face
- Lichenoid eruption and
- Hyperpigmentation over photo-distributed area
- Lymphoedema (acral)
- Vesiculobullous lesions
- Photo/ acral urticaria
- Multiple ecchymotic spots

## DISCUSSION

Chikungunya fever is caused by an arthropod borne alphavirus belonging to Togaviridae family and genus alphaviridae transmitted by vector Aedes mosquitoes (Aedes aegypti & A. albopictus). Chikungunya virus (CHIKV) can be both endemic and epidemic.[1] The name chikungunya came from the word 'Swahili' of MECONDE language, meaning "That which bends up". It was first described in 1955 by Marion Robinson & WHR Leumsdon and first reported from Tanzania in 1952 and in India from Kolkata in 1963.

Mother to child transmission is described although uncommon. During the epidemic peak in Reunion Island, the attack rate was as high as 8.3% in pregnant women.[2] The greatest risk of transmission is during birth if mother has acquired infection days before delivery.[3] CHIKV can

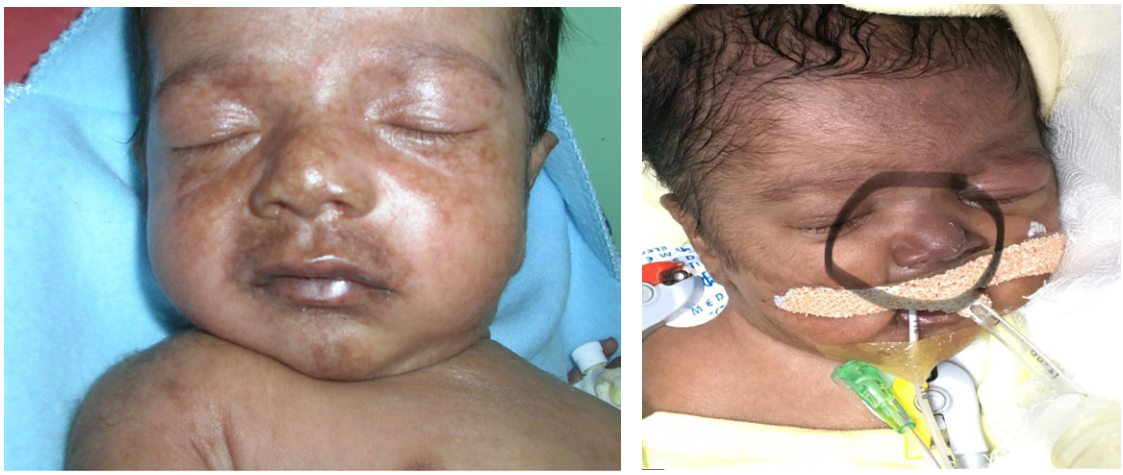

**Figure 1.** The characteristic 'CHIK SIGN'- Nasal blotchy erythema and freckled pigmentation over central face

be transmitted vertically with a probability of 50% when parturient women has a high viral load during the early stage of labor.[4,5]

The incubation period is 1-12 days after the mosquito bite. Initially high fever, followed by flu like symptoms are noticed. Joints of the extremities will be swollen and painful, hence the name Arthritic virus. Patients can develop Dengue & Chikungunya simultaneously. Features may be similar but less incidence of hemorrhage and shock in chikungunya. The clinical features may mimic septicemia due to bacterial infection, pyogenic meningitis or metabolic encephalopathy.

Neonatal infection could be associated with fever, poor feeding, tenderness, unexplained apnoea, distal oedema and various skin manifestations.[6] Patterns of skin involvement may include generalised pigmentation, macular rash, freckle like macules especially over face and nose and flagellate pigmentation over trunk and extremities.[7] The hyperpigmentation may develop soon after the rash has resolved and is a unique feature noted in Chikungunya. Hyperpigmentation is usually macular and most commonly affects nose and cheeks. Perioral, centrofacial hyperpigmentation (the chik sign) is a characteristic sign **(Figure 1).** Usually pigmentary changes develop two weeks after the rash. Hyperpigmentation may last for weeks to months. Post inflammatory hyperpigmentation could be due to virus triggered increased intradermal dispersion or retention of melanin **(Table 1)**.[8]

A higher incidence of encephalitis in neonates with Chikungunya could be attributed to greater viral replication and delayed clearance in infants.[9] A striking clinical feature is neurotropism manifesting as lethargy, refusal to feed and seizures. Neurological spread occurs through areas in brain poorly protected by blood brain barrier.[10] The possible mechanisms of neuronal damage include: invasion of choroid plexus and leptomeninges leading to defective neuronal migration. Another possibility could be microglial activation.[11] MRI brain may demonstrate hyper intensities on T2 and FLAIR images, involving frontal and parietal lobes in bilateral peri-ventricular and subcortical region with diffusion restriction due to cytotoxic oedema and capillary leak. Severe cases may show cystic encephalomalacia, ventricular dilatation or diffuse cerebral atrophy.

These cases are prone to developmental delay and require long term neurodevelopmental follow up.

## CONCLUSION

These two cases are being presented to create awareness of this condition where the baby can be severely affected with neurodevelopmental consequences. The early clues to diagnosis and avoidance of unnecessary investigations and treatment include taking a proper history, knowledge of the occurrence timing during the rainy season, involvement of family members with fever, joint pain and oedema or arthritis like picture and the typical hyperpigmentation (the CHIK-sign) in the infant**.** Knowledge of this cutaneous sign is extremely useful in a resource –poor setting to detect an unrecognized out break of Chikungunya so that adequate vector control measures can be adopted to prevent further spread of the disease.

There is no vaccine available to prevent chikungunya and hence prevention becomes important by reducing the number of mosquito breeding sites and by taking personal protection measures to avoid mosquito bites.

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