Benign Neonatal Sleep Myoclonus: Is it so Uncommon?

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Introduction

Benign neonatal sleep myoclonus (BNSM) is a self-limiting disorder characterized by neonatal onset myoclonic jerks during non-rapid eye movement (NREM) sleep, and consistent cessation with arousal with absence of concomitant electroencephalographic findings [1]. The importance of this entity lies in differentiation from other serious causes of neonatal seizures [2]. We report two cases with this condition in a span of one year which has been reported sparsely in literature.

Case Report

Case 1

A term female neonate, 3rd product of a nonconsanguinous marriage born by spontaneous vaginal delivery (birthweight-3.0kg) to a 28 year old mother with uneventful antenatal period, had a normal transition to postnatal life and was discharged on exclusive breastfeeds. She was brought on day 8 with a history suggestive of abnormal movements of limbs noted over the preceding 2 days. There was no other significant contributory history and the family history was negative for any seizure disorder. The baby was admitted with a diagnosis of neonatal seizures. Evaluation revealed a normal neonate with no abnormality on examination. In the hospital, the jerky movements lasted between 30 seconds to 3 minutes, mostly involving distal part of limbs without any change in vital parameters. However, as myoclonic jerks generally have a poor prognosis, a sepsis screen, metabolic profile, USS cranium, EEG and CT scan were carried out. All investigations were normal. The neonate had poor response to glucose, calcium, phenobarbitone and phenytoin. The baby continued to be active and feeding while the myoclonic jerks kept recurring. The jerks were then noticed to occur only during sleep and aborted during wakefulness. This finding helped us in establishing a diagnosis of BNSM. The jerks reduced in frequency from day 20 of life. Baby was sent home on tapering doses of anticonvulsant and was on fortnightly follow up. Anticonvulsants were stopped after two months. The myoclonic jerks disappeared completely by 4 months and baby was found to be normal.

Case 2

A term female neonate, first product of a nonconsanguinous marriage, born by caesarean section for transverse lie (birthweight 3.5kg) to a 25 year old mother with uneventful antenatal period was noted to have abnormal movements of limbs, predominantly distal, lasting for up to 1-2 minutes occurring during the sleep on day 3 of life while in hospital. The jerky movements disappeared when the baby was woken up. There was no other associated complaints and family history for seizures was negative. In this case, baby was observed for any variability in vital parameters or neurological abnormality. The neonate was diagnosed to have BNSM. She was discharged on breastfeeds with an advise to report in case of problem. On follow up, the jerks steadily reduced in frequency and disappeared by 5 weeks of life. Subsequent examination at 6 months revealed a completely normal infant.

Discussion

BNSM was first described by Coulter and Allen in 1982 [3]. Thereafter cases have been reported though it is likely that many cases of BNSM were managed with alternative diagnosis [4]. A transient serotonin imbalance apart from genetic factors may be involved in its etiology [5]. The myoclonic jerks in BNSM can be focal, multifocal or generalized. They characteristically appear only during NREM sleep and stop consistently when the child is aroused. This feature of BNSM helps in its recognition as well as its differentiation from neonatal seizures [1]. The majority of the neonates have generalized myoclonia, mainly in the distal part of limbs, jerking is of short duration, lasting 10 to 20 seconds though they may persist for more than 30 minutes [5,6]. The general condition of the child is unaffected and there are no associated abnormalities on neurodevelopmental examination [7]. The jerks of BNSM may be precipitated by stimuli such as sound or touch or even by benzodiazepines [1,3,4]. There are no associated EEG abnormalities in BNSM, which has been confirmed by long term video-electroencephalographic monitoring [8].

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An EEG may be required at times to distinguish BNSM from neonatal convulsion [1]. The jerky movements should be differentiated from jitteriness/clonus, which occur due to poor myelination of corticospinal tract in neonates. The closest alternative diagnosis is benign myoclonus of early infancy, which may rarely have a neonatal onset and have a normal EEG but these typically occur when the infant is awake. Others like benign familial neonatal seizures and benign idiopathic neonatal seizures (fifth day fits) are also included in the list of benign seizures of neonates and can be differentiated based on typical features. The rest of spectrum of neonatal seizures is related commonly to perinatal or metabolic insults, infections, developmental anomalies, drugs and hematological causes which can often be elucidated during evaluation [9]. BNSM is a self-limiting disorder with spontaneous resolution between 2 to 6 months of age and a normal neurodevelopmental outcome on follow up [1, 10]. Both the cases described fit into the above description of BNSM with typical myoclonic jerks occurring only during sleep and an entirely normal neurological status and outcome.

BNSM is frequently investigated and treated for seizures like in our first case [10]. This case report has been brought out to increase the awareness towards the same

Conflicts of Interest

None identified

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