Case Report

Can a Neonate Shiver ? — A perioperative Diagnostic Dilemma and a Short Literature Review

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Background: Neonates often show involuntary movements peri-operatively which are difficult to diagnose. Neonatal Shivering albeit very rare, may confuse the anaesthesiologist by presenting as Seizures.

Case : A Neonate was scheduled for the repair of Lumbar Meningomyelocele in our Operating Room. At the end of an uneventful Surgery, we noticed jittery movements during recovery from Anaesthesia. Differentiating these movements from Neonatal Shivering, Motor Automatisms and Physiological New-born Behaviour was a challenge. The immediate venous blood gas analysis was within normal limits and no apparent cause could be found. Suspecting accidental Hypothermia, Immediate rewarming was initiated and after Consulting Paediatric Neurology intravenous Levetiracetam was given. There were no further similar episodes.

Conclusion : Shivering-like episodes in Neonates during the peri-operative period need to be carefully evaluated to allay parental anxiety, avoid inappropriate treatment and prevent any further complications.

[*J Indian Med Assoc* 2022; **120(3):** 68-70]

Key words **: Neonatal convulsions, Benign, Shivering, Meningomyelocele, Hypothermia, MRI scan, Infant, Newborn, Diseases.**

Neonatal involuntary movements have a unique presentation, quite distinct from the older age groups. The accurate diagnosis of these movements is challenging especially if they occur during the peri-operative period, since multiple possible etiologies such as a Neurological Pathology, the surgical procedure itself or even the anaesthetic technique used should be considered. A thorough pre-operative evaluation and meticulous management during the peri-operative period is imperative. We present the case of a Neonate presenting for excision of a Lumbar Meningomyelocele who had jitteriness during emergence from Anaesthesia.

Written informed consent was obtained from the parents to publish this case.

Case Report

A 13-day-old Neonate with a lumbar Meningomyelocele (MMC) was scheduled for Surgical Excision of the MMC. The 2.9 kg female infant was born at 38 weeks' Gestation by Cesarean Section (due to non-progress of labour) to a 32-year-old primigravida whose pregnancy was complicated by Intrahepatic Cholestasis of pregnancy. The neonate had Appearance, Pulse, Grimace, Activity, Respiration (APGAR) scores of 8 and 9 at 1 and 5 minutes respectively and was alert, active and did not show any signs of neurological deficit. She was vaccinated with Bacillus Calmette Guerin (BCG), Oral Polio Vaccine and Hepatitis B Vaccine at birth and received an intramuscular Vitamin K injection. A Neurosurgery Consultation was done for the swelling over the lower back. On their advice a Magnetic Resonance Imaging (MRI) of the Spine was done which revealed a 5x4 cm Meningomyelocele at L5 Level with a Spina Bifida at L5-S1 Level and a low-lying Tethered Cord. The MRI brain was normal with no evidence of Tonsillar Herniation. The surgery was planned on day 13 of life. After an appropriate period of fasting, the baby was taken to the Operating Room (OR) and an intravenous induction was done using fentanyl, propofol and atracurium followed by securing the airway with a 2.5 mm Cuffed Endotracheal Tube with direct Laryngoscopy. After placing the neonate prone, Laminectomy of the L3-L4 spinous processes and excision of the MMC was performed. There was a small dural tear during the Surgery which was repaired. The intraoperative vitals were stable and there was minimal blood loss. Normothermia was maintained throughout the Surgery using a forced air warming device and warm fluids. The core temperature was monitored with a Nasopharyngeal Probe. We removed the probe a few minutes before extubating and the patient was exposed

Editor's Comment :

- Neonates may show abnormal movements in peri-operative period.
- The anaesthesiologist should be cautious to differentiate Shivering from Seizures or jitteriness and should intervene early to achieve better outcome.

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Received on : 30/12/2021
Accepted on : 24/01/2022

to the OR Temperature for a brief period. During recovery from Anaesthesia, the baby had Paroxysmal onset of jitteriness, clonic movement of the legs and the right upper limb with twitching of facial muscles (Supplementary digital content can be found in https://jimatube.in").

These movements could not be stopped by gentle physical restraint. We immediately covered the patient and started re-warming, following which the intensity and frequency of the movements diminished. The vitals were stable throughout this episode with no desaturation or Haemodynamic Disturbance. We took a trial of extubating after adequate recovery of muscle power and the Neonate maintained well on supplemental Oxygen by face mask. Oxygen saturation and respiratory efforts were maintained on room air trial as well. However, the Neonate did not cry and appeared sedated. These movements, albeit subtle by this time, continued shifting from one limb to the other at definite intervals. A venous blood gas analysis was done which showed a compensated Metabolic Acidosis (pH-7.36, pCO2-27 mmHg, pO2-41.9mmHg, HCO3-17.3 mmol L^{-1}). Electrolytes, Blood Glucose and Lactate Levels were normal (Na+-136 mmol L^{-1}, K+-5.1 mmol L^{-1}, Glucose-104 mg dl^{-1}, Lactate-1.3 mmol L^{-1}). As we could not ascertain whether these movements were due to Shivering, Seizures or any Physiological new-born behaviour, a Paediatric Neurology opinion was sought. The neonate was examined in the OR itself and Intravenous Levetiracetam 100 mg was initiated on their advice. The Neonate was then shifted to the Neonatal Intensive Care Unit and Intravenous Levetiracetam was continued in the postoperative period. The postoperative course was uneventful, and no further episodes of abnormal movements were noted.

Discussion

Neonatal Seizures have a distinct presentation, Pathophysiology, and Electroencephalogram (EEG) findings when compared to older age groups due to the Immaturity of the Neonatal Brain[1]. As a result, they can be a challenge to diagnose especially if they occur in the OR. Seizures occur due to the imbalance between the excitatory and inhibitory Neuronal Discharges within the Cerebral Cortex and can be precipitated by various factors such as Metabolic Disturbances, Intracranial Bleed, Infections, Congenital Brain Malformations etc. They Manifest in the Neonates as Clonic, Tonic or Myoclonic Seizures which can be focal or generalized[2].

Epileptic Seizures (movements that occur due to change in electrical activity in the brain) must be differentiated from normal Physiological New-born Behaviour that have a much better prognosis and often require no treatment. This is important so that appropriate treatment can be initiated where required and over-treatment avoided. These include Sucking movements, benign Neonatal sleep myoclonus, Neonatal tremors, Jitteriness etc[3]. Motor automatisms such as bicycling movements of the lower extremities, repetitive mouth and tongue movements, tonic posturing can be differentiated from Seizures by their ability to be triggered with tactile stimulation and inhibited by restraint. These are Non-epileptic, but a Neurology workup may be necessary if they are associated with other clinical features to rule out any Neurological issues[4].

Commonly used Anesthetic Agents can Precipitate Seizures especially during induction or emergence. Seizure-like phenomena with propofol have been widely reported while some other observations suggest that propofol may also have Anticonvulsant Properties[5]. Sevoflurane has also been shown to induce Epileptiform EEG and Seizure-like motor activity in children during anaesthetic induction and emergence[6].

There has been a reported increase in incidence of seizures 7 to 10 days after vaccination with Mumps, Measles, Rubella (MMR) Vaccine[7]. Although there is no consensus about the delay of Surgery following vaccination, Ingelmo *et al* recommend that Elective Surgery be postponed for a week after inactive vaccination and 3 weeks after live vaccination as Anesthesia and Surgery may cause immuno-suppression and may also cause confusion between vaccination side-effects and postsurgical complications[8]. The Neonate in our case had been vaccinated with BCG, Oral Polio Vaccine and Hepatitis B Vaccine at birth. Whether these vaccines also increase the risk of Seizures needs further evaluation.

The primary Pathology in this case was a Lumbar Meningomyelocele and 17% of children with Meningo-myelocele have associated Seizure Disorders. Most of these children have underlying Central Nervous System (CNS) Pathology to account for their Seizures such as Cerebral Cortical Dysplasia, Polymicrogyria and Cortical Dysgenesis[9]. However, the pre-operative MRI in our case did not show any evidence of brain pathology.

A few other differentials were also considered in our case. Collins *et al* reported 2 cases of jitteriness among New-borns where the only Biochemical Abnormality that could be found was Vitamin D deficiency. They suggested that Neonatal Tremors could be an early indicator of Vitamin D deficiency and should be considered in the work up of Neonates with no other known Pathology[10].

Neonates predominantly maintain Normothermia using Non-shivering Thermogenesis although Brück et al. have noted that Shivering has been observed in Neonates with Severe Hypothermia after Birth[11]. Alexander *et al* suggested that shivering might occur if Non-shivering Thermogenesis had reached its full potential[12]. Propofol, fentanyl and inhalation agents have been shown to reduce Non-shivering Thermogenesis in Infants and could be an inciting factor in triggering Shivering in

Neonates[13]. Maintaining Normothermia is crucial in the perioperative period and this is especially true for Neonates who have underdeveloped Thermoregulatory Systems. On the other end of the Temperature Spectrum, Febrile Convulsions are a Common Cause of Seizure Disorder in Children with most of them occurring within 24 hours of fever onset[14]. The warming blanket used during anaesthesia can cause an increase in body temperature which itself can cause Seizures in a Neonate[15]. In our case, the small duration for which the Neonate was not covered with the warming blanket, could result in Hypothermia. Thus, Neonatal Shivering was considered as a differential diagnosis.

Conclusion

Neonatal Seizures or seizure-like Phenomena during the perioperative period can be challenging to diagnose and manage, often leading to an increased duration of postoperative stay, increased parental anxiety and at times inappropriate treatment. One must consider all possible factors related to the patient, surgery and anaesthesia while searching for the etiology. Neonatal shivering, although rare, should be kept as a differential diagnosis of these abnormal movements.

Conflict of interest : The authors declare no conflict of interest.

Funding : Nil.

Acknowledgement : Dr Sourav Ray (MBBS) for editing the video clip.

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