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Perioperative Anesthetic Management in Preterm 12 Days Infants with Giant Meningoencephalocele at Mid Occipital and Secondary Trigonoccephaly

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Abstract

Introduction: A meningoencephalocele is herniation of neural element along with meninges through a congenital defect in cranium. The incidence of encephalocele is approximately 1/5000 live births; occipital encephalocele is more common in females than males. It is called as giant meningoencephalocele when the head is smaller than the meningoencephalocele. These giant meningoencephaloceles harbor a large amount of cerebrospinal fluid (CSF) and brain tissue, so there occur various surgical challenges and anesthetic challenges in positioning and intubation. **Case:** A 12 days neonate was consulted to the neurosurgery department with complaints of large swelling over the back of head and difficulty in feeding. She was diagnosed with ventriculomegaly and meningoencephalocele since 32-33 pregnancy. The swelling was small at the time of birth, but it gradually increased in size. The child was born by section caesarean because of fetal distress and meningoencephalocele. The neonate current weight was 3.195 grams with Post Conceptional Age (PCA) 35-36 weeks. On examination, the patient large spherical swelling was present over occipital region and there was no head control. The patient was active, conscious with no impression of focal neurological deficit. Systemic examination was unremarkable. The head circumference was 30 cm and circumference of occipital swelling was 40 cm. Potential problems in this patient include preoperative preparation and optimization of general condition, difficulty in positioning the patient, difficult airway (intubation), periodic apnea and potential hemodynamic disturbances and a sudden decrease in intracranial pressure during cele resection. **Conclusion:** Perioperative management in this case started from preoperative to postoperative evaluation. Preoperative preparation in anticipation of airway difficulties and communication with the operator is very important. Appropriate anesthetic techniques should aim to maintain stable hemodynamics and oxygenation and prevent a sudden increase or decrease in intracranial pressure.

Keywords: Airway, Infants, Intracranial Pressure, Meningoencephalocele, Perioperative Management

1. Introduction

Encephalocele is a rare lesion, being an embryological mesodermal anomaly which results in a defect in the cranium and dura, associated with herniation of meninges, cerebrospinal fluid, or brain tissues through a defect

usually covered by scalp. Surgical management of children with giant occipital encephalocele requires careful attention to pediatric anesthetic and surgical principles (Ganeriwal et al., 2019). Meningoencephalocele is a type of Encephalocele. It's an abnormal leakage of cerebrospinal fluid (CSF) and herniation of brain tissue and meningeal membranes through a defect in the bony skull. It's a rare congenital condition and occurs approximately in 1 per 5000 live births (Senapathi et al., 2021). It is categorized into two types according to the sac's locations: occipital and frontoethmoidal. Multiple theories have been proposed to explain the exact cause of Meningoencephalocele. Many observations and associations have been found to be co-occurring with Meningoencephalocele. Despite that, there is no single theory that clearly explains the pathogenesis of this anomaly. Therefore, it's believed to be due interaction between genetic background and environmental factors. Meningoencephalocele is diagnosed antenatally using sonography. It can achieve diagnostic accuracy in 80% of cases. Other imaging modalities including: CT scan, MRI, and MRA can also be used for further detailed evaluation but their use has been limited due to the rarity of this anomaly. Several factors influence the prognosis of patients with Meningoencephalocele. The sac size and the amount of herniated brain tissue determine the prognosis. In addition, hydrocephaly, infections, and other anomalies accompanying the condition determine the prognosis as well. The mortality rate approaches 30% despite the applied appropriate treatments (Senapathi et al., 2021). We present a case of a giant occipital encephalocele in 12 days neonates highlighting the anesthetic problems encountered in its perioperative management.

2. Case Report

A preterm 12 days neonate with 3,195 gram bodyweight was consulted for elective cele – resection surgery. She was admitted to the High Care Neonatal Unit (Neonates HCU) following a section caesarean labor because of fetal distress and meningoencephalocele. His Apgar score at 1 min was 5, and 9 at 5 min. She had 43 cm body length and 31 cm of head circumference, within normal parameters. Since he was born, his parents noticed that there was a lump on the back of his head. The lump is big as Takraw ball which could be bigger when he was crying. At HCU, the patient was kept in a prone position and nil per oral. Examination showed an active baby moving all limbs normally with no neurological deficit. There is no periodical apnea although he was preterm neonates with 35 weeks of PCA. Neurosurgical team was consulted and they recommended Computerized Tomography (CT) scan and Magnetic Resonance Imaging (MRI). They also recommended intubation should any problem arises. A provisional diagnosis of Giant Meningoencephalocele was made.

Upon taking a detailed history from the mother, she was 34 years old, Gravida 3 Para 2+ 1. The mother attended once in the last trimester of her Antenatal Care visits for throughout this pregnancy because of traveling a lot due to personal reasons. Her past obstetrical history was a spontaneous abortion one and half years back. Her other children are all healthy and were delivered vaginally with no similar complaint. When his mother at 32-33 weeks of pregnancy, she was brought to Fetomaternal Department at Hasan Sadikin Hospital, underwent Fetomaternal USG, diagnosed with ventriculomegaly and meningoencephalocele, she was hospitalized for 10 days and plan to do caesarean section. Upon reviewing the CT scan and MRI reports, the neurosurgical team recommended surgery because The sac has the potential to rupture and cause significant CSF leakage and a sudden decrease in intracranial pressure. This has the potential to cause death, although there was no brain tissue within the herniated sac.

On physical examination, the neonate consciousness was fully alert with Children Coma Scale (CCS) 11, pulse rate 145-150 beats per minute, respiratory rate 43-45 times per minute without periodic apnea, temperature 36.9°C and oxygen saturation 98% with free air. On examination of the head, there was a mass in the occipital area measuring 40 cm in diameter, soft consistency, with the impression of fluid content. The conjunctiva was not anemic, the sclera was not icteric, the pupil is diameter of 3 mm both eyes with the both light reflexes were positive. On examination the airway was difficult to assess. Neck examination revealed limited range of movement (ROM). Neurological examination revealed no motor weakness, visual impairment was difficult to assess. On thoracic examination, the shape and motion were symmetrical, the first and second heart sounds were regular, murmurs and gallops were absent, the both lungs had the same breath sounds, and there were no additional breath sounds in both lungs. Abdominal examination revealed a flat abdomen, not tense, normal palpable liver/spleen, normal bowel sounds. On examination of the extremities found warm sensation with capillary refill time less than 2 seconds.

On the laboratory findings, We found the results of: Haemoglobin: 12.6 g/dl, Haematocrit: 37.7 %, Leukosit count 16.5300 / μ L, Platelets: 757.000 / μ L, Albumin: 3,09, Sodium 128 mEq/L, Potassium: 4,9 mEq/L, the others parameters of laboratory and chest x-ray were in normal range. An MRI examination revealed a meningoencephalocele with a defect in the right occipital bone with suggestive of corpus callosum colpocephaly. On head ultrasound there was the fluid structure with cerebral parenchyma appears in the occipital area. There is no hyperechoic appearance of the subarachnoid. No calcification was seen in the lateral periventricular white matter or in the cerebral cortex with the impression of meningoencephalocele. Other investigations were within normal limits.

The Diagnosis was Meningoencephalocele at Mid Occipital with Ventriculomegaly, Secondary Trigonocephaly synostosis and Preterm Infant 33-34 weeks.

Picture 1: Head CT Scan and MRI Images

Picture 2: Chest X-Ray Image

Picture 3: Head USG Image

3. Anaesthetic management

A day before surgery the patient was planned to fast 6 hours before surgery. Maintenance fluid was given with 0.9% NaCl 12 ml/hour. Pre Surgery, the patient was in active movement, alert and hemodynamic was within normal limits. During Surgery, hemodynamic monitoring using Non Invasive Blood Pressure (NIBP) measurement, pulse oximeter, capnography monitor and thermometer. The patient is positioned on his side with a circular support pad for the meningocele segment. The patient was preoxygenated with 100% O₂. Then performed induction with spontaneous breathing technique using volatile gas sevoflurane. After the patient was asleep, the patient's position was changed to lie flat with a back cushion so that the head and body form a straight axis.

Picture 4: Clinical picture during pre-induction with modified support pad (lateral position)

Picture 5: Clinical picture during induction with modified support pad (supine position)

Picture 6: Clinical picture after induction with lateral position

Intubation was performed using a video laryngoscope with a 2.5 non-cuff endotracheal tube (ETT). After intubation, fentanyl 15 mcg and atracurium 2 mg were given. The ETT was fixed and an intraoral packing gauze was applied. The depth of anesthesia was maintained with sevoflurane 3-4 vol% with 50% oxygen. Breathing support was fully controlled by manual ventilation with Jackson reese neonate circuit. The surgery taken were cele resection and dura mater suturing. Total blood loss was 25 cc and being replaced by transfusion of 20 cc of Packed Red Cell (PRC). During intraoperative hemodynamic was in stable condition.

After the surgery was over, hemodynamic and respiratory assessments were performed. The patient was extubated. Then the patient was transferred to the semi intensive-neonatology care unit.

4. Discussion

Anesthesia management in neurosurgery cases begins with the principles of neuroanaesthesia that have been applied during perioperative anesthetic management. Management of the patient's airway is kept securely at all times, by doing pre-induction positioning during surgery, breathing is controlled by manual bagging with the installation of a capnograph with the aim of maintaining EtCO₂ levels of 25-30 mmHg and preventing hypoxia starting from preoperative with oxygen use, preoxygenation for 5 minutes prior to single-step intubation. The patient's hemodynamics was maintained by close observation of the pulse, intraoperative bleeding and communication with the operator regarding CSF drainage. The depth of intraoperative anesthesia is maintained so that the patient does not wake up because it will cause secondary brain injury. PaO₂ levels during surgery are maintained at 100-200 mmHg to prevent cerebral vasodilation at PaO₂ levels < 50 mmHg.

In cases of occipital meningoencephalocele; herniation of meninges, occipital lobes, and/or ventricles are common. Other contents of meningoencephalocele may be cerebellum, brainstem, or rarely, torcula. Torcula as one of the contents of encephalocele poses a greater challenge as its injury may lead to cerebral deep venous system thrombosis and its associated consequences of assault to the already compromised brain (Pahuja et al., 2015). In this case, the sac wasn't contained brain at all, its composed of liquor cerebrospinalis and also segment of duramater. But its also have a significant risk of being drop of volume of LCS because of leak and rapid aspiration. The rapid decrease of LCS may cause herniation of brain and hemodynamic disturbances (Bisri et al., 2019; Jacob et al., 2008).

Preoperatively, preparation for significant blood loss should be made because of potential bleeding from the suboccipital bone and the dural sinus. The ultimate prognosis, however, depends on various factors. Proper positioning of the patient is required for successful endotracheal intubation. Although, usually, patients with an occipital meningoencephalocele are operated in prone position, a giant size prevents this positioning and the patient has to be kept in lateral position as seen in the present case. Endotracheal intubation may be difficult due to large swelling and short neck, so alternative airway management options should be kept ready before starting anaesthetic induction. In this case we use support bearing on the back and round support pillow below the meningocele. This support aimed to make horizontal line between body and head during supine position just before intubation. For emergency alternate way we prepared laryngeal mask airway of appropriate size, with high-frequency jet ventilation, fibre optic bronchoscope, a cricothyroid cannula, and preparations for tracheostomy should be made. Due to a low functional reserve volume, neonates are more prone to develop hypoxia, hypotension, and bradycardia and even cardiac arrest. Therefore, very close monitoring is required. Elective surgery provides the time to patients to gain weight and strength and offers the surgeon for selection of the best technique. However in this case, giant meningoencephaloceles require urgent surgical treatment to avoid damage to sac. The leak of sac was avoided because it may cause rapid drainage of LCS and being life threatening. In this surgery, communication with neurosurgeon is important to make sure there was a slow drainage of LCS to avoid rapid decrease of intracranial pressure. There was aspiration of the CSF before skin incision in large meningoencephalocele helps in dissection of the sac. In this case of circular meningoencephalocele with a small occipital bone defect, a transverse incision was made. Care should be taken to identify the contents of the sac. Rarely, the sagittal sinus torcula and the transverse sinus are seen in the vicinity of the sac. It is desirable to preserve the neural tissue. The dura has to be repaired meticulously to get a water-tight closure to prevent CSF leak. Many factors affect the outcome of patients with occipital meningoencephaloceles which include site, size, amount of brain herniated into the sac, presence of brainstem or occipital lobe with or without the dural sinuses in the sac,

and presence of hydrocephalus. The presence of gross brain tissue in sac, associated hydrocephalus, or congenital anomalies is unfavorable prognostic factors. In this case we found no brain segment in the sac and also there was no sinus involvement in the sac. This is very beneficial because it avoids secondary brain damage, herniation, massive and uncontrolled and bleeding.

Conclusion

The management of occipital meningoencephalocele can be complicated and should be individualized. In a tense, giant occipital meningoencephalocele problems encountered are essentially because of the large size and induced neonate handling, positioning in operation theatre, intubation, and blood loss during resection of the large amount of redundant skin. we must always carry out close observation with communication with the operator being the key to safety in this surgery.

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