



# Anaesthetic managements of 16 days' neonate with large occipital meningoencephalocele in a resource-limited setting, Ethiopia: a clinical case report and review of literature

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**Introduction:** Meningoencephalocele is a rare congenital neural tube defect that results in herniation of brain tissue, necessitating surgical treatment. However, anaesthetic management of meningoencephalocele is challenging because of the giant occipital mass in airway management, particularly for anaesthetists working in resource-limited settings. Therefore, this case report aimed to share the challenges encountered during the perioperative anaesthesia management of a giant occipital meningoencephalocele in Ethiopia.

**Case presentation:** A 16-day-old female neonate was referred from a health centre in Ethiopia for surgical excision and repair of a giant occipital meningoencephalocele (GOM). The main challenges were hemodynamic instability and airway management due to the giant mass on neck movement and inability to achieve the optimal position for intubation.

**Methods:** A single clinical case report discussed challenges encountered during perioperative anaesthesia management of GOM in a resource-limited setting in Ethiopia, and this work has been reported in line with the SCARE 2023 criteria.

**Conclusion:** Anaesthesia management in neonates with GOM is challenging for anaesthetists in resource-limited settings because of the scarcity of different airway equipment and large masses. Another concern was to avoid pressure on the GOM due to rupture, which may result in hemodynamic instability and hypothermia. Therefore, attention should be paid to proper handling of the airway, hypothermia, and fluid loss. Generally, managing an occipital meningoencephalocele poses challenges for anaesthetists, particularly in terms of securing the airway. Despite these challenges, the authors managed with locally available resources.

**Keywords:** airway challenge, case report, hemodynamic instability, occipital encephalocele

## Introduction

Meningoencephalocele is a rare congenital condition characterized by protrusion of neural tissue, which is caused by failure of the neural tube to close completely during foetal development<sup>[1]</sup>. Additionally, it is characterized by sac-like protrusions that cover the brain and membranes through openings in skull defects<sup>[2,3]</sup>. The prevalence of encephalocele varies among different regions of the world, with 70% of cases being occipital<sup>[4,5]</sup>. In Africa 2 cases per 10 000 births<sup>[6]</sup>, In Ethiopia 1.07 cases per 10 000 births<sup>[7]</sup>.

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## HIGHLIGHTS

- Notable positioning challenges were more prominent in cases of giant meningoencephalocele (GOM).
- Intubation can be performed in the supine position with two assistants supporting the head and a giant cyst/mass.
- Successful management of neonates with GOM requires consideration of a combination (preventing hemodynamic fluctuations and excessive pressure on the sac to avoid rupture, and managing a possible difficult airway due to the mass and restricted neck movement).
- Healthcare providers working in resource-limited areas may need to adopt unconventional methods based on available resources and setup to prevent further complications.
- It is better to manage early with available resource, as a delay in the surgical management may lead to adverse outcomes, specifically in resource-limited countries.

Paediatric airway management in the presence of a giant occipital meningoencephalocele (GOM) can be challenging because of difficulties in positioning, mask ventilation, and intubation<sup>[4]</sup>. This challenge is greater in developing countries<sup>[8]</sup>. Positioning and airway management in any patient with occipital encephalocele are challenging, particularly for anaesthetists working in resource-limited areas. The scarcity of airway

equipment creates optimal conditions for tracheal intubation by providing airway alignment and stability without increasing pressure on the encephalocele. Hence, one of the primary goals of perioperative management of GOM is gentle manipulation to prevent rupture and secure the airways.

Positioning the patient's head beyond the operating table for intubation contributes to undesirable head movements during laryngoscopy and difficulties in airway management. Lateral positioning is less ideal approach for the laryngoscopist and potentiates an obscured laryngoscopic view<sup>[9,10]</sup>. Removal of encephalocele contents by needle aspiration before intubation may potentiate infection, hemodynamic instability, and possible herniation<sup>[9]</sup>. To overcome these challenges, anaesthetists have developed innovative ideas using locally available resources and techniques to manage the airway<sup>[11]</sup>.

In this case, tracheal intubation in the lateral position was difficult. The patient was then shifted to the supine position after a failed attempt (two trials) in the lateral position and shifted to the supine position for tracheal intubation using a doughnut shape prepared from packs and drapes with locally available materials. Therefore, this clinical case report aimed to highlight the anaesthetic challenges encountered and to discuss the methods used to overcome with locally available resources. This study was reported in line with the SCARE 2023 criteria<sup>[12]</sup>.

### Case presentation

A 16dayold female neonate presenting with giant cystic swelling (giant meningoencephalocele) in the occipital region was referred from a district hospital to the University of Gondar Referral Hospital after delivery via caesarean section. The neonate was a pre-term (37 weeks) birth weighing 2.9 kg with Apgar scores of 7 and 8 at the first and 8 min, respectively, with no evidence of family history, and the mother had no prenatal or antenatal care (ANC). The patient was scheduled for surgical excision and repair, and admitted to the paediatric unit in our hospital for preoperative evaluation by anaesthetists; each system was normal except for physiological jaundice and redness at the site of the sac (swelling measuring 15 × 11 cm) arising from the occipital region, and larger than that of the neonate head) (Fig. 1).

On examination, the cardiovascular auscultation (S1 and S2 well heard), the respiratory rate was 37/min with equal air entry bilaterally, oxygen saturation (SpO<sub>2</sub>) on room air was 96%, and temperature was 36.7°C. Upon investigation, complete blood count (CBC), electrocardiography (ECG), abdominal ultrasound (AU), and electrolyte levels were normal. No other congenital anomalies were noted. After written informed consent was obtained from the mother of the neonate and transferred to the operation theatre (OR) for surgery.

### Intraoperative management

Standard ASA monitoring was applied with anaesthetists supporting giant cysts in the operating room (OR), and premeditated with fentanyl (1 µg/kg) to suppress airway reflex, atropine 0.02 mg/kg for bradycardia, and calculated maintenance fluid continued. Preoxygenation was performed by mask ventilation with 100% oxygen. The mass was gently handled using locally prepared doughnut-shaped ring along the horseshoe, prepared from a pack of appropriate size, was kept under the back,



**Figure 1.** An image shows neonate with large occipital meningocele after intubation.

whereas the other two anaesthetists, and neurosurgeon avoided pressure over the sac in the supine position. This was followed by administration of ketofol (1:1) for induction.

The supine position was considered after a failed trial of tracheal intubation in the lateral position and desaturation was observed. Due to the lack of video laryngoscopy, the miler blade, and the GOM on neck movement made challenging the face mask ventilation and intubation in the lateral position (lateral attempt was failed); then, we shifted to supine position for intubation and intubated. In the second attempt in supine position, the trachea was intubated using an uncuffed 3.0 mm inner diameter (ID) endotracheal tube (ETT) with a stylet inside and confirmed with direct vocal view using a laryngoscope along auscultation in the supine position confirmed/successfully intubated the patient. All laryngoscopy attempts were quick, and no desaturation was noted; then the patient was shifted to the prone position for surgery, followed by confirmation of appropriate ETT placement at a depth of 9 cm (Fig. 2).

The intraoperative monitoring shows: heart rate of 150 beats/min, blood pressure (BP) of 65/37 mmHg, and SpO<sub>2</sub> of 96%. Balanced solutions with 1% dextrose and 6 ml/kg/h, along with boluses of isotonic fluids (10 ml/kg), were used. Despite this, persistent hypotension occurred during the surgery, and blood product transfusion was initiated according to the maximum allowable blood loss. Intraoperative anaesthesia was maintained using 1–2% isoflurane and oxygen. Intraoperatively, the heart rate dropped to 70 beats/min with hypotension; atropine and a fluid bolus of crystalloids, and blood were then administered.

In total, 260 ml of fluid infusion (100 ml of blood and 160 ml of normal saline) was administered.



**Figure 2.** Neonate with giant occipital meningocele after intubation shifted to prone position for surgery.

### **Outcome and postoperative course**

After surgery, the patient transferred to the ICU as intubated to observe smooth and spontaneous recovery in the ICU, and extubation was performed after establishment of spontaneous respiration, and recovery on the day of the surgery. Maintenance fluid, oxygen and 80 mg paracetamol was initiated. The neonate had a smooth postoperative course, except for delayed recovery from anaesthesia, which may be due to the intraoperative complications reported, and relieved on the postoperative course. The patient was closely followed up for complications. However, the postoperative course was uneventful, and the patient was discharged after smooth recovery on the seventh postoperative day.

### **Case discussion**

Perioperative management of patients with GOM may be challenging for both anaesthetists and neurosurgeons. Perioperative management of GOM includes proper positioning without pressure to prevent cyst rupture. Another concern is subsequent positioning during the procedure. The meningoencephalocele may also associated with other varying degrees of congenital defects, and restricted neck extension, making intubation difficult<sup>[13,14]</sup>. However, in this case, we did not find any abnormalities other than GOM.

Perioperative complications commonly encountered during anaesthetic management of GOM include bradycardia, hypoxaemia, and bronchospasm<sup>[15-17]</sup>. Another study also reported a higher incidence of latex allergy, manifesting as intraoperative bronchospasm and cardiac collapse<sup>[15,16]</sup>. Once the decision to operate has been made, a perioperative plan must be formulated

by an anaesthetist based on airway management, fluid balance, and hypothermia prevention<sup>[5,10]</sup>. Anaesthetic management in these children requires monitoring of body temperature, and estimation and replacement of blood loss. Latex allergy precautions also should be considered with these high risk procedure<sup>[1,10]</sup>.

The positioning problem is more prominent in cases of large occipital encephalocele<sup>[13]</sup>. In addition, one study reported higher difficulty in airway management in children with congenital malformations<sup>[4,11]</sup>. Alternative methods for successful intubation include rolled-up blankets placed under the head of the child in the lateral position or needle decompression of the encephalocele sac under sterile conditions<sup>[9]</sup>. Intubation can be attempted in the supine position, with an assistant supporting the head. The baby can be placed on a pile of blankets, and the sac is protected in a doughnut shape with a drape, pack, and support by an assistant. NMB agents should only be administered after checking for adequate mask ventilation. In this case, we managed the airway by placing the neonate in a supine position, and the sac was protected by elevating the head with a pack of assistants for intubation<sup>[4,17,18]</sup>.

### **Strengths and limitations**

In this case report, we successfully managed the case, specifically the airway, by placing the neonate in a supine position using a locally available and prepared by anaesthetists. Difficulty in the lateral position, and highlighting the importance of considering the adoption of unconventional equipment with the available resources and setup with open mind to manage and prevent further complications as a delay in the surgical management may lead to adverse outcomes, specifically in resource-limited countries. There may be a referral bias before the patients presented to our hospital for surgery. Despite this limitation, we highlighted the challenge and importance of adopting unconventional airway equipment with the available resources and setup, along with a review of literature.

### **Conclusion**

In conclusion, the key to the successful management of neonates with GOM is a combination thus attention should be paid to intraoperative blood loss/preventing hemodynamic fluctuations, and along perfect handling of the airways with proper positioning. Hence, it poses the challenge of preventing excessive pressure on the sac to avoid premature rupture and to manage a possible difficult airway due to the mass. The challenges encountered were important; thus, difficult intubation in the lateral position in a resource-limited setting due to lack of advanced airway equipment and the need to adopt unconventional with the available resources and setup, but safe ideas for better management with successful outcomes.

The patient was discharged after a smooth recovery on the seventh day of the surgery, with instructions for follow-up to observe the progress, and no complications were reported. Further studies are required to better understand this, as this was a single case.

## Ethical approval

The case reports were exempted from ethical approval at our institution.

## Declaration of patient consent

Informed consent was obtained from the neonate's mother, including images (Fig. 1), for publication in a journal for educational purposes. The mother understands that their names and initials will not be published and due efforts will be made to conceal their identity; however, anonymity cannot be guaranteed. Data were obtained through direct observation and management of patients during perioperative anaesthesia. A copy of the written consent form is available for review by the Editor-in-Chief upon request.

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No funding was received for this study.

## Author contribution

The corresponding author observed perioperative anaesthesia management, wrote the case report, prepared the manuscript, selected the journal, and sent for publication. All authors have read, wrote, and approved the case reports.

## Conflicts of interest disclosure

The authors declare no conflict of interest.

## Research registration unique identifying number (UIN)

None.

## Guarantor

The corresponding author (A.F.I.).

## Data availability statement

All the data used in this study can be accessed through direct communication with the corresponding author.

## Provenance and peer review

Not commissioned, externally peer-reviewed.

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