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Case Report

Anaesthesia Section

Perioperative Anaesthesia Challenges in a Difficult Airway Case of a Turricephaly Child Posted for Fronto-orbital Advancement and Remodelling: A Case Report

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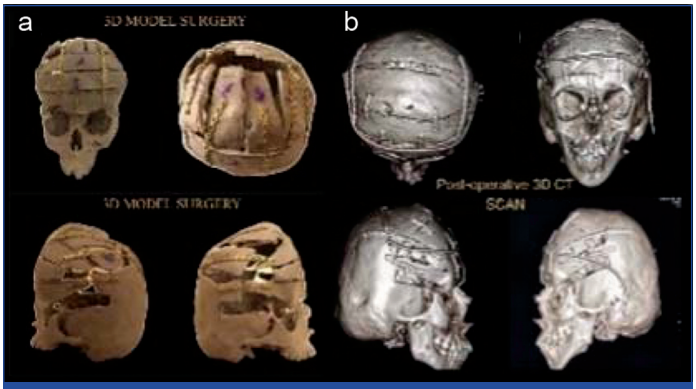
ABSTRACT

Turricephaly is a rare craniosynostosis characterised by premature fusion of the coronal sutures, resulting in a tower-shaped, elongated skull. This abnormal skull morphology presents anaesthetic concerns due to associated craniofacial deformities that complicate airway management, increase the risk of raised intracranial pressure and pose challenges in vascular access, fluid management and control of intraoperative blood loss. Present case is of a 13-year-old girl with turricephaly who underwent fronto-orbital advancement and cranial remodelling. An anticipated difficult airway was encountered, requiring multiple intubation attempts. Intraoperative anaesthesia care focused on securing the airway, maintaining normocapnia, invasive monitoring, proper fluid management, prevention of hypothermia and management of intraoperative blood loss with transfusion of blood products. Postoperatively, the patient was electively intubated and transferred to the Paediatric Intensive Care Unit (PICU) and kept on a ventilator overnight with appropriate sedation and analgesia. The patient was extubated the next day. This case highlights the importance of thorough preoperative assessment, vigilant intraoperative management including fluid management and transfusion strategies, maintenance of normothermia and meticulous postoperative care in achieving a successful outcome in such complex craniofacial surgical cases.

Keywords: Craniosynostosis, Skull and face surgery, Congenital anomaly, Skeletal deformity

CASE REPORT

A 13-year-old female, weight 25 kg, height 140 cm, presented with turricephaly (fronto-occipital flattening with a tower-shaped skull) skeletal deformities scheduled for fronto-orbital advancement and remodelling. The patient was evaluated with general and systemic examinations and screened to rule out other congenital anomalies. A complete blood count, coagulation profile, renal function tests, liver function tests, 3D-CT scans of the craniovertebral junction, MRI brain with venography, Electrocardiography (ECG), chest X-ray and 2D echocardiography were performed. A 3D model was printed and the surgeons preplanned the sections on it, enabling the surgery to be performed in a planned, stepwise manner [Table/Fig-1]. The patient's baseline Haemoglobin (Hb) was 11.3 g/dL. Estimated Blood Volume (EBV) and Maximum Allowable Blood Loss (MABL) were calculated to be 1625 mL and 200 mL, respectively [1]. Packed Red Blood Cells (PRBCs), Fresh Frozen Plasma (FFP), and Platelet Concentrates (PCs) were readily available. The patient had micrognathia, a deviated tongue and crowding of teeth, but a mouth opening of 38 mm [Table/Fig-2]. This case required proper parental counselling and familiarity with the child to maintain calm. Informed written consent (assent) for anaesthesia {American Society of Anaesthesiologist Grade III}, for central venous and arterial lines, for multiple blood transfusions, and for postoperative elective ventilation was obtained from the parents. Indirect Laryngoscopy (IDL) with a 90° video laryngoscope showed bilateral mobile vocal cords. An otorhinolaryngologist was kept on standby for an emergency tracheostomy. With a multidisciplinary approach, a team of oral and maxillofacial surgeons, anaesthesiologists, neurosurgeons, paediatricians and nursing staff was dedicated to this case perioperatively to yield a successful outcome.



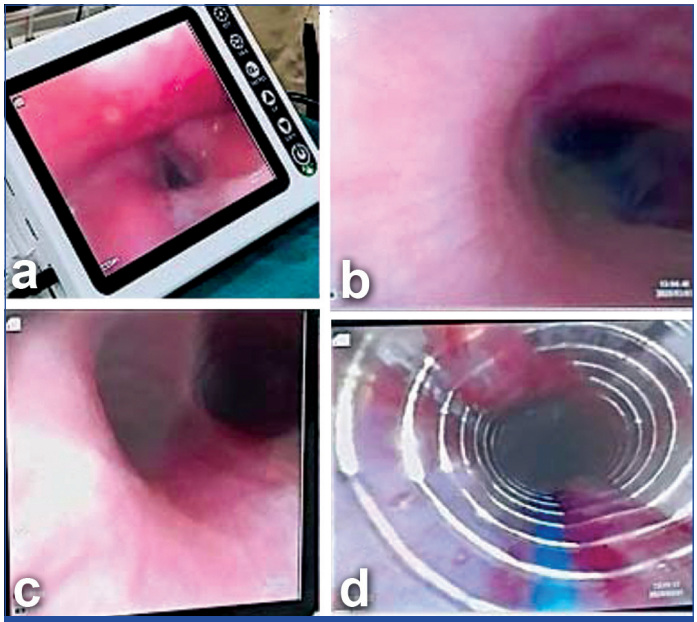
[Table/Fig-1]: a) Preoperative 3D modelling of the skull; b) Postoperative 3D CT scan of the skull.

[Table/Fig-2]: a) Airway assessment; b) Preoperative; and c) Postoperative profiles of the patient.

Intraoperative management: The patient was brought to the ENT operating theatre with a 20-G intravenous (i.v.) cannula in the left hand. A warm-air blower blanket was placed to maintain body temperature. Maintenance fluids were started as Ringer's lactate. Eight parameters were monitored, including ECG for Heart Rate (HR),

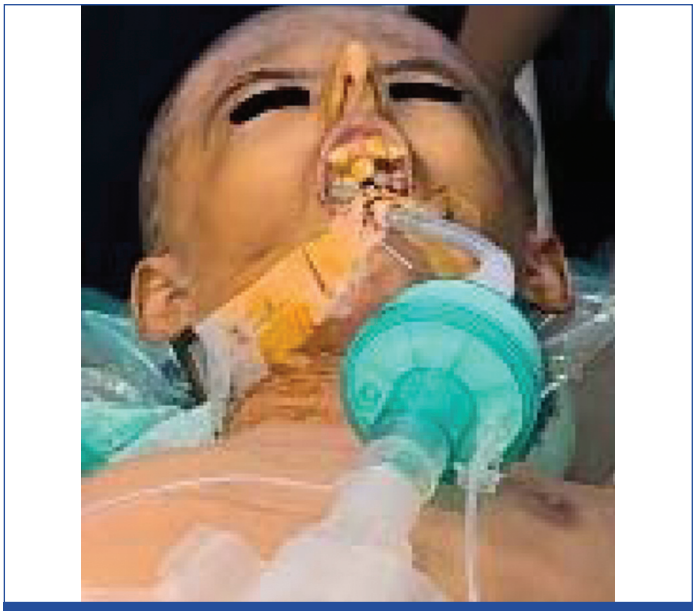
Non invasive Blood Pressure (NIBP) for Systolic BP (SBP), Mean Arterial Pressure (MAP), and Diastolic BP (DBP); Pulse Oximetry (SpO_2); End-Tidal CO_2 ($ETCO_2$); Invasive arterial Blood Pressure (IBP); Central Venous Pressure (CVP); Respiratory Rate (RR); and a temperature probe (T). Premedication included intravenous glycopyrrolate 0.1 mg, ondansetron 2.5 mg, paracetamol 240 mg, dexamethasone 8 mg, and hydrocortisone 50 mg i.v.

An adequate pillow was placed under the head to provide a sniffing position for laryngoscopy [2]. The patient was preoxygenated with 100% Oxygen (O_2) for three minutes. Induction was achieved with intravenous propofol 70 mg and maintained with 100% O_2 plus sevoflurane on spontaneous ventilation, with brief attempts not exceeding 20 seconds. With adequate mouth opening, paediatric The Anaesthesia Society (TAS) scope was used for intubation. Despite clear visualisation of the vocal cords, the first two attempts at intubation aided by a bougie failed due to the lack of a bougie guide channel slot in the TAS scope. Subsequently, another anaesthesiologist attempted with a channelled video laryngoscope, but it was also unsuccessful. An attempt with the Video Bronchoscope (VB) also failed. Oxygen was provided through the oral cavity via an open circuit at 8 L/min to provide passive oxygenation. After every unsuccessful attempt, the patient was given 100% O_2 with sevoflurane to avoid hypoxia and to maintain depth of anaesthesia. On the final attempt with the VB, the vocal cords were visualised [Table/Fig-3]. Intravenous succinylcholine 50 mg was administered, and intubation was successfully performed by advancing a 5.0 mm internal-diameter armoured reinforced cuffed endotracheal tube over the VB. After direct visualisation of the Endotracheal Tube (ETT) placement above the carina, the cuff was inflated and the VB was gently removed. Thereafter, the ETT was secured in the midline by the surgeons with sutures and wire fixation and elastic adhesives [Table/Fig-4].



[Table/Fig-3]: Vocal cords visualisation and Video Bronchoscope (VB) guided intubation: a) Vocal cords; b) Trachea; c) Carina; d) ETT in situ.

Anaesthesia was maintained with low-flow anaesthesia using a 50% O_2 and air mixture at a flow of 1 L/min, sevoflurane at 1 Minimum Alveolar Concentration (MAC), with the help of an Anaesthesia Gas Monitor (AGM). Fentanyl 50 µg plus 25 µg and an atracurium loading dose of 12 mg were given, followed by continuous infusion at 12 mg/h. The patient was ventilated in Pressure-Controlled Ventilation (PCV) mode with a Tidal Volume (TV) of 200 mL (approximately 8 mL/kg), respiratory rate 22/min, Post End Expiratory Pressure (PEEP) 4 cmH_2O, plateau pressure 15 cmH_2O, and peak inspiratory pressure 16 cmH_2O to minimise barotrauma and maintain normocapnia (ETCO2 35-45 mmHg). Temperature was monitored with a rectal probe and maintained at 36.6-37.0°C (98.0-98.6°F) using a warming



[Table/Fig-4]: Midline fixed ETT with suture, wires and sticking.

blanket. A throat pack was placed to prevent spillage into the glottic area. Antibiotic coverage was cefoperazone-sulbactam 1.5 g IV and amikacin 500 mg IV after a test dose. Right Internal Jugular Vein (IJV) cannulation was performed using a 5.5 Fr triple-lumen catheter under ultrasound guidance with CVP monitoring. Radial artery cannulation could not be secured due to wrist deformity; therefore, right ulnar artery cannulation was performed using a 22-G IV cannula for arterial blood pressure monitoring. Thereafter, the patient was catheterised for urinary monitoring. Intraoperative blood loss was approximately 500 mL and was replaced with PRBCs totaling 545 mL (243 mL and 302 mL), one unit FFP 228 mL, and one PC 72 mL. Mannitol 10 g IV and frusemide 10 mg IV were given for diuresis, and calcium gluconate 50 mg IV was administered. Baseline Arterial Blood Gas (ABG) prior to transfusion and post-transfusion ABG and venous samples were collected [Table/Fig-5]. The patient remained haemodynamically stable throughout, with SBP maintained between 90 and 110 mmHg. At the end of surgery, intravenous levetiracetam 500 mg was given for antiepileptic prophylaxis. The patient was electively shifted to the PICU with the ETT in place. The total duration of anaesthesia was seven hours.

Postoperative management: In the PICU, the patient was kept on Pressure-Regulated Volume-Controlled (PRVC) ventilation mode, with VT 200 mL, RR 18, FiO2 0.5, PEEP 4 cmH_2O, maintaining an ETCO2 of 27 mmHg. Intravenous 0.9% normal saline 500 mL plus Potassium Chloride (KCl) 10 mEq at 50 mL/h as maintenance fluid was given. Analgesia and sedation were managed using infusions of dexmedetomidine (2 µg/mL), 5 mL/hour, and fentanyl (10 µg/mL), 2.5 mL/hour for 14 hours. Additionally, diclofenac 1 mg/kg and paracetamol 5 mg/kg twice daily were administered. To reduce facial oedema, a tapering regime of dexamethasone was started for three days: 1 mg TID, BID, and OD. For cerebral protection to reduce intracranial pressure and to avoid seizure activity, mannitol 20% 50 mL TDS and levetiracetam 10 mg/kg BD were started respectively. Serial ABG analyses were planned to guide ventilation, and venous samples were collected for routine investigations to correct Hb, electrolytes, or infections. Diuretics and calcium gluconate were given as per the anaesthesiologist's advice due to blood transfusions. Continuous monitoring of HR, IBP, RR, SpO_2, and $ETCO_2$ was performed. Postoperative Hb was 8.9 g/dL. An additional PRBC at 15 mL/kg was transfused to improve Hb [Table/Fig-5]. The patient was extubated 17 hours after surgery following a satisfactory T-piece trial and ABG analysis. A repeat 3D-CT scan was performed on first Postoperative Day (POD) [Table/Fig-1]. The patient was shifted to the ward on the third POD and discharged on POD-13 [Table/Fig-2].

Arterial blood gas (ABG)				Venous blood gas (VBS)				
	Intra op.	POD 0	POD 1		Pre op.	Intra op.	POD 1	POD 2
pH (7.35-7.45)	7.4	7.51	7.57	INR (0.8-1.2)			1.6	1.27
pO_2 mmHg (75-100)	290	237	238	CRP mg/L (<10)	2.32	2.32	28.7	77.8
pCO_2 mmHg (35-45)	42	37	26	Platelets lacs/mm^3 (1.5-4.5)	4.5	3.42	3.9	2.6
HCO_3 mmol/L (22-26)	26	29.5	23.8					
Hb g/dL	7.8	9.9	8.1	Hb g/dL	11.3	8.6	8.9	12.1
Ht % (35-45)	25	32	24.3	Ht % (35-45)	33.9	25.8	26.7	36.4
Lactate mmol/L (0.5-1.4)	1.5	3.0	0.8	S. creatinine mg/dL (0.3-0.7)	0.6		0.5	0.5
Ca^{++} mmol/L (1.16-1.31)	1.02	1.04	0.96	Ca^{++} mmol/L (1.16-1.31)				2.25
$S.K^+$ mmol/L (3.5-5.5)	4	2.7	2.8	$S.K^+$ mmol/L (3.5-5.5)	4.1	4.1	2.9	2.9
$S.Na^+$ mmol/L (135-145)	137	139	138	$S.Na^+$ mmol/L (135-145)	140	137	133	138
PRBC[§] (1 Unit=300 mL)						2	1	
FFP[†] (1 Unit=200 mL)						1	1	
PC[‡] (1 Unit=70 mL)						1	2	

[Table/Fig-5]: Perioperative ABG analysis and VBS.
[§]Packed Red Blood Cell (PRBC); [†]Fresh frozen plasma, [‡]Platelet Concentrates (PC)

DISCUSSION

Turricephaly is a rare congenital cranial deformity resulting from premature closure of the coronal or multiple cranial sutures, leading to increased cranial height and a tower-like skull shape. It is reported to have an estimated incidence of 1 in 100,000 to 200,000 births, with a male predominance (male-to-female ratio 3:2) [3,4]. Overall, craniosynostosis affects approximately 1 in 2,000 to 2,500 live births [5]. Turricephaly poses several anaesthetic challenges, including difficult airway management, risk of raised intracranial pressure, abnormal facial anatomy, intravenous access difficulty, temperature regulation issues and the potential for significant intraoperative blood loss [6]. Surgical procedures like fronto-orbital advancement involve extensive bone manipulation, further increasing anaesthesia risk.

Turricephaly presents multifaceted perioperative challenges, the most critical being airway management. The goals for this surgery were: (i) to maintain a patent airway and successful intubation; (ii) to maintain Hb, blood viscosity and coagulation profile; (iii) to maintain normocapnia and normothermia; (iv) to avoid volutrauma and/or barotrauma to the lungs. A stepwise approach underscores the importance of preparedness and flexibility in managing difficult airways, especially in craniofacial deformities. However, intubation via direct laryngoscopy is successful in the majority of cases [7]. Video laryngoscopes are designed for patients with difficult intubation. Studies show that these devices improve the Cormack–Lehane laryngoscopic view and can be used to aid endotracheal intubation [8]. Despite a modified Mallampati Grade II airway, this patient had failed intubation attempts, reinforcing the notion that external craniofacial features often do not reflect internal airway complexity [9]. A guarded approach for induction with propofol and sevoflurane under spontaneous respiration ensured the patient's safety in case of failed intubation attempts. The manoeuvrability of video bronchoscopy through an oral pathway poses challenges to its use compared with the natural curvature during nasal bronchoscopy [10]. Only when the vocal cords were visualised, a depolarising muscle relaxant was used to prevent obstruction due to cord movements; this proved effective in this case. In patients with a challenging airway, spontaneous ventilation should be sustained until the airway is adequately secured [11]. Given the nature of the surgical procedure and the relative difficulty in accessing the airway, it is paramount to ensure that the ETT is not prone to kinking or compression. This can be achieved by using a reinforced ETT and, most importantly, securing it to prevent accidental dislodgement, which could result in either endobronchial intubation or accidental extubation. Methods to secure the ETT include adhesive tape, sutures and wiring [7].

Fluid and blood loss management is pivotal, especially in surgeries involving large cranial flaps and osteotomies. Surgeries lasting more than five hours are associated with high blood loss. There are critical phases of surgery when the blood loss is expected to be greatest. Understanding these events, along with the surgeon, enables the anaesthesiologist to reduce them. Most of the blood loss occurs during scalp incision and detaching the periosteum [12]. Infiltration with 2% lignocaine with adrenaline 1:200,000 can reduce blood loss during scalp incision. Blood loss in such cases can exceed 20-30% of EBV [13]. Autologous transfusion preoperatively is preferred in such cases, but due to time constraints, donor blood transfusion was arranged after crossmatching, including PRBCs, FFP and platelets. A preplanned calculation of Allowable Blood Loss (ABL) and its corresponding cut-off limit helps maintain Hb, blood viscosity and coagulation [14,15]. It is imperative to know the normal haematocrit and blood volume to calculate ABL. There are several methods for estimating blood loss, such as gravimetry, photometry and serial haemoglobin measurements. These methods have limited use in practice because of unavailability and time consumption. Hence, visual estimation—using mops, gauze and drapes—is the most commonly used method for estimating blood loss [1]. The transfusion trigger has been described as Hb level of 7-8 g/dL or haematocrit of 27-30% [12]. Normal blood volume based on age and gender for this patient was 65 mL/kg [1]. The most common indication for blood transfusion is the "10/30" rule, which states that Hb should be maintained at or above 10 g/dL or haematocrit at or above 30%. Calculation of ABL: ABL=EBV×(Hi – Hf)/Hi, where EBV is the estimated blood volume and Hi and Hf are the initial and final haematocrits, respectively; EBV=weight (kg)×blood volume per kg (age and gender) [1]. Postoperative blood loss is generally due to oozing from large areas of exposed bone surface [16]. Haemostatic blood products can be infused empirically when blood loss exceeds 1.5 times the calculated ABL, although this depends on protocols at different centres [12].

Patients undergoing extensive perioperative transfusions may develop coagulopathy due to blood loss, haemodilution, insufficient component replacement and consumption of coagulation factors. The conventional approach to managing a severely bleeding patient involves restoring normovolaemia by volume replacement concurrently with addressing anaemia and coagulopathy. Timely therapeutic intervention in surgical bleeding is paramount to prevent compromised haemodynamics, diminished oxygen-carrying capacity secondary to anaemia and impaired coagulation haemostasis resulting from depleted haemostatic resources [14].

In this case, the goal was to maintain the preoperative Hb and provide optimal fluid management to maintain viscosity and avoid

haemodilution. Arterial line catheterisation was essential for beat-to-beat blood pressure and heart-rate monitoring, cardiac output monitoring [17] and periodic ABG analyses to assess Hb and electrolyte status, which aided decisions regarding blood transfusion management alongside visual blood loss assessment [14,18]. The arterial line was kept through POD 1 to monitor hypotension and low Hb levels due to oozing; it can be maintained through POD 3 [19]. A FloTrac system that calculates cardiac output from arterial-line data was unavailable. This device is easy to implement as it does not require central venous or pulmonary artery catheterisation or transpulmonary thermodilution techniques and it helps in implementing haemodynamic management protocols [17]. Given skeletal deformities posing difficulties with peripheral access, the IJV was essential for large fluid shifts, periodic sampling, anaesthetic management and potential inotropic support, with consideration of risk of Venous Air Embolism (VAE) and blood transfusions [15,20].

Temperature control is critical, as hypothermia (<36.0°C) is common due to prolonged surgery and blood exposure [21]. Intraoperative warming and temperature monitoring helped maintain normothermia, reducing the risk of coagulopathy [21,22]. Besides anaesthetic actions, hypothermia also impairs platelet and coagulation function and increases transfusion requirements [21]. It is advisable to maintain core body temperature >36.0°C with the help of fluid-warming devices and forced-air warming from the beginning of the surgery, which was done in this case [21]. ABG monitoring ensures continuous assessment of gas delivery, oxygenation, MAC, $ETCO_2$, and circuit integrity in low-flow anaesthesia [23,24]. VAE is a known complication of such surgeries, reported in up to 83% of cases, usually without haemodynamic compromise; clinically significant VAE occurs in about 1-2% [20]. It can be detected with precordial Doppler; $ETCO_2$ monitoring can also aid detection.

The patient was kept on PCV intraoperatively. Due to skeletal deformities, there is a tendency toward a restrictive ventilation pattern. PCV is advantageous in such patients because it caps peak airway pressures, avoiding barotrauma or volutrauma and promoting even distribution of ventilation. It enhances alveolar recruitment, reduces Ppeak and improves the ventilation/perfusion ratio [24].

Given a prolonged surgery (>7 hours), anticipated craniofacial swelling and an established difficult airway, combined with the complex nature of strip craniectomies, it was a joint decision by the surgeon and anaesthesiologists to keep the patient electively on ventilation in the PICU under multidisciplinary care. Most patients are extubated at the end of surgery. Factors that may delay extubation include a prolonged procedure, marked fluid shifts, large-volume transfusions, the effects of prolonged prone positioning and patient factors such as preoperative obstructive sleep apnoea or airway concerns [20].

Postoperative care included ventilatory support (PRVC mode) and multimodal analgesia with dexmedetomidine and fentanyl infusions, promoting haemodynamic stability, analgesia and early extubation with ABG monitoring, along with antiepileptic coverage and steroids to reduce inflammation and swelling [25-27]. Effective analgesia improves neurological recovery, reduces metabolic demand and lowers the risk of postoperative complications [25]. Analgesia was provided as multimodal therapy, i.e., fentanyl, diclofenac, paracetamol, and dexmedetomidine, administered intraoperatively and postoperatively. Drain outputs and serial ABG measurements guided therapy. Analgesia in the first 24 hours is typically intravenous opioids, with a transition to an oral regimen by 24-48 hours [12]. ABG analysis helps optimise ventilation and extubation decisions. A P/F ratio >200 is generally considered for extubation [28].

The patient remained on PRVC mode with sedation overnight. Sedation was discontinued in the morning to assess spontaneous respiration. A T-piece trial was performed; after satisfactory respiration, extubation was performed. A multidisciplinary approach is essential for optimal outcomes. Preoperative planning, anticipation of complications and readiness for escalation of care can significantly influence prognosis in such complex craniofacial reconstructions.

CONCLUSION(S)

Turricephaly presents several challenges for the anaesthesia team, particularly in managing a difficult airway, significant blood loss and maintaining stable vital signs during prolonged surgery. Thorough preoperative planning, the use of advanced airway equipment, close monitoring with invasive lines and effective fluid and temperature control are crucial. Postoperative care, including appropriate sedation and pain relief, facilitates a smoother recovery. A team-based approach is paramount to safely manage such intricate cases and achieve optimal outcomes.

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