

Management of Anticipated Difficult Airway in A 5 - Year - Old Child with Embryonal Rhabdomyosarcoma of the Uvula in a Resource Poor Environment

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ABSTRACT

Rhabdomyosarcoma is a rare tumour of the oral cavity, with the embryonal subtype being the most common. It typically presents in children under the age of 10 and has a predilection for the head and neck region, although involvement of the oral cavity is less frequent. Airway management in paediatric patients presents inherent challenges due to the unique anatomical and physiological characteristics of this age group. These challenges are further compounded in cases where a difficult airway is anticipated. To ensure patient safety and effective airway management, thorough history-taking and physical examination are essential, particularly in patients with noticeable anatomical abnormalities of the airway. Close collaboration between the anaesthesia and surgical teams is crucial to ensure that all relevant clinical concerns are identified, understood, and addressed. This collaboration facilitates comprehensive preoperative planning, especially when managing anticipated difficult airways. We present the case of a 5-year-old boy with residual embryonal rhabdomyosarcoma of the uvula, scheduled for debulking surgery. Airway management proved challenging, requiring two intubation attempts and a modification of the initial technique. The surgery and postoperative recovery were uneventful. This case highlights the practical application of conventional techniques in managing paediatric difficult airway cases within a resource-limited setting and underscores the importance of adaptability and multidisciplinary planning.

Keywords: Difficult airway management, embryonal rhabdomyosarcoma, laryngoscopy, paediatric.

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INTRODUCTION

Embryonal rhabdomyosarcoma is the most common subtype of rhabdomyosarcoma, accounting for approximately 49% of all cases. [1,2] It typically occurs in children under 10 years of age, with the highest incidence observed in those below 4 years. While it is occasionally seen in adolescents and young adults, younger age at diagnosis—particularly below 10 years—is associated with a better prognosis. [3] Despite its relative frequency among soft tissue sarcomas, embryonal rhabdomyosarcoma is a rare condition, representing approximately 3% of childhood cancers and 2% of cancers in adolescents. [4]

The most frequently affected sites include the head and neck region (notably the extraocular muscles of the eye), the genitourinary tract, and the retroperitoneum. Within the head and neck region, less commonly affected sites include the nasal cavity, nasopharynx, ear, paranasal sinuses, and soft tissues of the face and neck. Oral cavity involvement—such as the tongue, lip, and palate—is rare. [5,6] The estimated incidence is about 4 cases per million children. [3] Risk factors associated with the development of embryonal rhabdomyosarcoma include fetal exposure to radiation, parental drug use, a family history of rhabdomyosarcoma in first-degree relatives, preterm birth, use of fertility drugs, and the presence of congenital anomalies. [7] Genetically, embryonal rhabdomyosarcomas have been linked to aneuploidies involving the RAS and TP53 genes. [7]

Airway management is a fundamental and often life-saving aspect of anaesthesia practice. Difficulty in securing the airway, or failure to do so, is a well-documented cause of anaesthetic-related morbidity and mortality. [8] This risk is heightened in paediatric patients due to the unique anatomical and physiological characteristics of this population. An understanding of these differences is crucial for safe and effective airway management. Paediatric airway techniques and equipment often differ significantly from those used in adults. However, in resource-

limited settings, the availability of specialised paediatric equipment may be limited.

The presence of pathological processes affecting the airway—such as tumors—can be predictive of a difficult airway. [9] In such cases, it is essential to maintain spontaneous ventilation during airway management, either through titrated doses of intravenous induction agents or via inhalational agents, until the airway is successfully secured. Initial attempts should be made using conventional laryngoscopy, with anaesthesia deepened adequately to facilitate the procedure. The number of laryngoscopy attempts should be kept to a minimum to avoid trauma, bleeding, and airway oedema [10]. If standard techniques are unsuccessful, clinicians should be prepared with an alternative airway management strategy, including the option of awakening the patient or proceeding to a surgical airway if necessary.

Case Presentation

A 5-year old male, weighing 15 kg, presented with difficulty in breathing which was worse while asleep, snoring and muffling of his voice. He was an old patient of the managing unit who had presented 2 years earlier on account of an intraoral mass affecting the uvula, for which he had examination under general anaesthesia and biopsy of the lesion. A subsequent histology report was in keeping with a diagnosis of embryonal rhabdomyosarcoma. The patient was immediately scheduled for tumour debulking surgery.

There was no history of asthma, sickle cell disease or seizure disorder and there was no history of food or drug allergies. He had no loose or missing teeth. There was no history of anaesthetic complications, nor transfusion reaction. He had received 13 cycles of chemotherapy 9 months prior to re-presentation, including doses of Vincristine, Actinomycin D and Cyclophosphamide.

Physical examination revealed a calm afebrile child who was not pale, anicteric, acyanosed and not

dehydrated, but had generalized lymphadenopathy and no pedal oedema. The pulse rate was 112bpm, respiratory rate was 20cycles per minute, pulse oximetry showed an oxygen saturation (SPO₂) of 98% in room air and temperature was 36.2°C.

Airway examination showed no craniofacial anomaly. There was a mobile irregular mass in the oropharynx attached to the uvula [Figure 1]. The mass was central, not erythematous and had greyish patches. The interincissor gap was 6cm and mouth opening was adequate. The thyromental distance was 4cm, sternomental distance was 12cm and the mallampati score was 1. There was adequate neck extension, the chest was clinically clear and the cranial nerves were intact.

Investigation

SEUCr – Na – 125mmol/l	K ⁺ – 4.3mmol/l
Cl ⁻ – 88mmol/l	HCO ⁻² – 25mmol/l
Urea – 1.8mmol/L	Cr – 4.9μmol/L
FBC – PCV – 34%	Hb conc – 11.4g/dl
Platelet count – 544 x 10 ⁹ /l	WBC – 6.49 x 10 ⁹ /l
PT/INR – 12.9 (9 -11) sec	INR – 1.0 (0.8 – 1.2) sec
PTTK - 30.5 (24 – 36) sec	LFT- normal

SEUCR: Serum electrolytes, urea and creatinine, K⁺: potassium, Cl⁻: chloride,

HCO⁻²: bicarbonate, Cr: creatinine, FBC: full blood count, Hb: hemoglobin, WBC: white blood cell count, PT/INR: prothrombin time/International normalised ratio INR: International normalised ratio, PTTK: partial thromboplastin time with kaolin, LFT: liver function test.

Histology reported a malignant mesenchymal neoplasm composed of small, stellate, spindled or round cells with minimal or moderate deeply eosinophilic cytoplasm and eccentric nuclei. The cells were disposed in sheets with hypo and hypercellular areas within a myxoid matrix. Strap cells and tadpole cells were also seen.

CT-Scan of the head and neck showed an uvula mass measuring 4.23cm x 2.37cm x 2.35cm with ill-

defined margin obscuring the airway. [Figures 2 and 3]

An assessment of American Society of Anesthesiologists (ASA) class II and residual embryonal rhabdomyosarcoma of the uvula was made. A unit of whole blood was grouped and crossmatched. The parents were counselled on the anaesthetic technique and written informed consent was obtained. The patient was fasted according to the standard fasting guideline, intravenous access was secured on the morning of surgery and IV fluid commenced.

In anticipation of difficult airway management, preparation for difficult airway management was made, a difficult airway tray set up and preparations possible emergency tracheostomy was made.

In theatre, anaesthetic machine and ancillary equipment check was done for availability and functionality, a difficult airway tray was prepared and emergency drugs were drawn and labelled. The patient was transported in supine position on a gurney, he was placed on the operating table, standard monitors were attached and baseline vital signs documented.

The patient was preoxygenated for 5 minutes and anaesthesia was induced using intravenous (IV) propofol 1.5mg/kg, test ventilation showed easy mask ventilation with adequate chest rise. Laryngoscopy was attempted using a size 3 Miller laryngoscope blade, for possibility of intubating without muscle relaxation but was unsuccessful. Visualization of airway structures was in keeping with Cormack and Lehane 4 (neither the glottis nor the epiglottis was visible) and the mass was completely obliterating the oropharynx.

Manual ventilation was immediately commenced with 100% oxygen to SPO₂ of 100%, anaesthesia was deepened with IV propofol 0.5mg/kg, IV atropine 0.015mg/kg and suxamethonium of 1mg/kg was administered. A second attempt at laryngoscopy revealed Cormack and Lehane grade 3 (tip of the epiglottis). With the use of a paediatric gum elastic bougie, he was successfully intubated by railroading

a size 4.5mm ID endotracheal tube and the tube was secured at 14cm mark.

Findings during surgery revealed a friable papillomatous soft palatal mass occluding the esophageal isthmus with destruction of the uvula. A debulking of the mass was done. He was scheduled for chemotherapy.

DISCUSSION

Embryonal rhabdomyosarcoma affecting the palate mostly involve the soft palate, and few of them involve the uvula. [11]

A difficult airway is the clinical situation in which anticipated or unanticipated difficulty or failure is experienced by a physician trained in anesthesia care, including but not limited to one or more of the following: facemask ventilation, laryngoscopy, ventilation using a supraglottic airway, tracheal intubation, extubation, or invasive airway.[12] Adverse outcomes of unsuccessful airway management include death, hypoxic brain injury, cardiopulmonary arrest, airway trauma, aspiration of gastric contents, pulmonary oedema and dental damage.[13] The catastrophic complications that ensue with failure of oxygenation or maintenance of a patent airway justify the numerous developments, research, guidelines and algorithms developed in this area. Advances in the delivery of airway training and education reinforce the need to learn to deal with this challenge. [14]

Difficult airway management is often anticipated in some patients. Risk assessment to predict a difficult airway may involve the evaluation of information from a patient's history or medical records. Such information includes data on demography, clinical conditions, diagnostic tests, patient/family interviews or questionnaires and physical examination. [12] Airway examination identifies the presence of upper airway pathologies or anatomical anomalies. Anatomical measures including Mallampati and modified Mallampati scores, thyromental distance, sternomental distance, interincisor distance, neck circumference, ratio of neck circumference to thyromental distance, ratio of

height to thyromental distance, hyomental distance, and hyomental distance ratio are useful in identifying patients that may possibly pose challenges during airway management. [12] In our study, an airway examination was conducted to identify anatomical anomalies. In addition, measurement of the interincissor gap, mouth opening, thyromental distance, sternomental distance and the mallampati score was used in assessing the airway for ease of endotracheal intubation. There was adequate neck extension, the chest was clinically clear and the cranial nerves were intact.

Airway complications are more frequent in patients with difficult airways, however, such cases do not occur frequently. [15] In an austere environment such as ours, the management of an anticipated difficult airway can present a challenge, given the limited availability of specialized airway equipment and facilities. The anaesthetist must have a good understanding of the anatomical and physiological anomalies present in the patient and possess adequate skill to safely manage and secure the airway. A thorough and meticulous preanaesthetic evaluation and investigation of the patient, a carefully drawn anaesthesia plan and interprofessional team cooperation is imperative for safe management of patients and securing good outcomes. Proficiency in difficult airway management may be acquired through simulation exercises and patient management over time.

In addition to clinical assessment, advanced diagnostics are useful in refining airway assessment and formulating precise airway management strategies. In our environment, X-rays are commonly used in assessing airway structures. With Computed Tomography (CT) scans, finer details of the anatomy can be seen. Magnetic Resonance imaging (MRI) better defines soft tissue lesions, such as invasive cancers and can be used to formulate airway strategies in tumours in and around the airways. CT scans and MRI are available in our environment, however, these facilities are expensive. Our patient had a CT Scan done, this was of immense value in

developing an airway management plan. Three-Dimensional printing (3D printing) simulates the airway model and enables planning, practice and education.[14] However, as at the time of carrying out this procedure the, the biomedical engineering department in our hospital did not have this equipment.

Fibreoptic direct laryngoscopes offer improved illumination. With video laryngoscopes, manipulation of the neck can be limited, lesions in the airway may be better appreciated and images of the airway can be recorded. This equipment was not available in our facility as at the time of carrying out this procedure. Patients at risk of airway collapse during induction may benefit from the use of awake fibreoptic bronchoscopy. Paediatric gum elastic bougies can be lifesaving in paediatric patients with poor Cormack and Lehane scores (3 and 4), as was the case in our patient.

In the course of managing our patient, we adopted a team-based approach, that involved a pre-induction briefing to discuss the anticipated airway challenges and management plan. This was done in line with the American Society of Anesthesiologists' and the Difficult Airway Society's practice guidelines for the management of difficult airways.

Researchers have explored the outcomes of use of different types of laryngoscope blades. Levering laryngoscope blades have been compared to the English Macintosh blade. [16] Although no difference in laryngoscopic view was found, shorter intubation times and fewer intubation maneuvers occurred with the use of the levering laryngoscope. In our case, we used a size 3 straight blade laryngoscope. Its slim and slender blade allowed ease of intubation without tissue trauma or debris dislodgement into the airway. Gentle maneuvering of the equipment also contributed to the success of our technique.

Some workers have demonstrated success with the use of combination techniques in securing the airway. Wu et al [17] compared the use of a

combination of a lightwand and with direct laryngoscopy to use of lightwand alone for intubation and reported equivocal findings for successful intubation, first attempt success, time to intubation and number of intubation attempts.[18] On the contrary, we employed a single technique given the constraint of limited availability of advanced equipment.

The 2022 ASA practice guidelines for management of the difficult airway in patients who can be adequately ventilated but difficult to intubate recommends that the anaesthetist identifies a strategy for successful airway management in such patients.[12] In our case, we decided to attempt laryngoscopy following intravenous induction without muscle paralysis as a primary plan with the view to subsequently paralyse the patient and repeat the attempt at intubation if the first technique failed. We achieved success with the later technique. The guideline also recognized that airway management in the uncooperative or paediatric patient may require an approach that may not be considered as a primary approach in a cooperative patient. This was the case in our patient.

It is important that the risks and benefits of awake extubation versus deep extubation be considered in such patients. There is need for the formulation of a plan of action in case of failure of extubation. While deep extubation may reduce the risk of laryngospasm or bronchospasm, coughing or bucking prior to removal of the tube, there is a risk of loss of pharyngeal muscle tone. Our patient was extubated awake because of his age and the need to make it easier to manage any inadvertent post-extubation complications.

The ASA recommends the placement of a stylet-type device or bougie through the endotracheal tube (ETT) and removing the ETT over the stylet. [10] Airway Exchange Catheters (AECs) which are long hollow semi-rigid catheters have been designed for this purpose. They allow for insufflation of oxygen and sampling of end tidal carbon dioxide.

Unfortunately, these devices are scarcely available in low resource centres like ours.

Post-extubation care in Intensive care or recovery room is important as patients may be at risk of airway obstruction within this period. Oxygen supplementation and monitoring should be continuous through the period of transportation and recovery, as was the case in our patient. The postoperative period was uneventful. In line with the American Society of Anesthesiologists' recommendations for the use of post-extubation steroids and / or racemic epinephrine when appropriate, our patient received intravenous dexamethasone 0.2mg/kg. [12] He was discharged from the recovery room upon satisfaction of the recovery room discharge criteria.

There is need for routine airway management training and simulation for anaesthetists as a means of acquiring proficiency in difficult airway management. Recently in Nigeria, the Society of Specialists in Airway Management (SSAM) has organized series of airway management workshops for doctors and other health workers. This has been followed by the establishment of airway simulation centres in various tertiary health facilities in the various geopolitical zones of the country. This has been contributory to training of anaesthesia manpower in airway management in the country.

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Author contributions

Example: IOO, conceptualized and designed the study. IOO, POO, FEM and EM contributed to implementation of the project, IOO and POO contributed to the revision of the manuscript. All authors were involved in the writing and revision of the manuscript. The authors read, approved the final manuscript and agree to be accountable for all aspects of the work.

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