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Letter to Editor

Bifid epiglottis: Managing the unforeseeable

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Dear Editor

Difficult airway is a taxing situation where an experienced anaesthesiologist experiences difficulty or failure with one or more of the following: facemask ventilation, laryngoscopy or tracheal intubation, extubation or an invasive airway.[1] Such situations are mostly the consequence of anatomical airway variations or abnormalities. Using these physical variations and attributes, difficult airway can be predicted. However, anatomical abnormalities involving trachea and larynx are frequently asymptomatic and can remain unsuspected until encountered during intubation.

Bifid Epiglottis (BE) is an extremely rare laryngeal anomaly, seen in neonates.[2] It is mostly associated with a syndromic picture and anomalies like polydactyly or genitourinary and cardiac abnormalities.[3] The child may present with aspiration or airway obstruction.[4] Some are even associated with laryngeal cyst and cricoid stenosis. Bifid epiglottis is an most commonly associated with syndromes such as Pallister-Hall Syndrome, Bardet-Biedl Syndrome, and Joubert Syndrome.

A 1-month-old female weighing 3kgs, with vaginal atresia and hydrometrocolpos was posted for vaginostomy. Birth history revealed preterm vaginal delivery. Physical examination revealed polydactyly of all the limbs. There were no features of facial dysmorphism. Her preoperative blood investigations were within normal limits. 2D echo was suggestive of a small atrial septal defect. USG KUB was suggestive of left sided hydroureteronephrosis.

In the operating room, standard ASA monitors (ECG, NIBP, and SpO2 probe) were applied. A 22-gauge cannula was already secured in the right hand, and Ringer's lactate was initiated as maintenance fluid. The child was premedicated with 30 micrograms of glycopyrrolate and 6 micrograms of fentanyl. Induction was performed with 6 mg of propofol and 1.5 mg of atracurium, followed by 3 minutes of ventilation.

The first attempt at laryngoscopy was made using a Miller size1 blade, but the glottic opening could not be visualized. A second attempt was made by a senior anaesthetist using the same laryngoscope, but again, the glottic aperture remained undetectable. A supraglottic device (I-gel size 1) was then inserted, but only marginal ventilation was achieved despite repositioning the device. Oxygen was administered via nasal prongs at 3 L/min before proceeding with the next attempt.

The subsequent attempt was made using a video laryngoscope with a size 0 blade. With external manipulation, the glottic opening was successfully visualized, revealing a bifid epiglottis (**Figure 1**). An endotracheal tube (ETT) with a 3 mm internal diameter was introduced and secured at 9 cm after confirming bilateral air entry. Throughout all attempts, the patient was ventilated with 100% oxygen, maintaining a saturation of > 95%.

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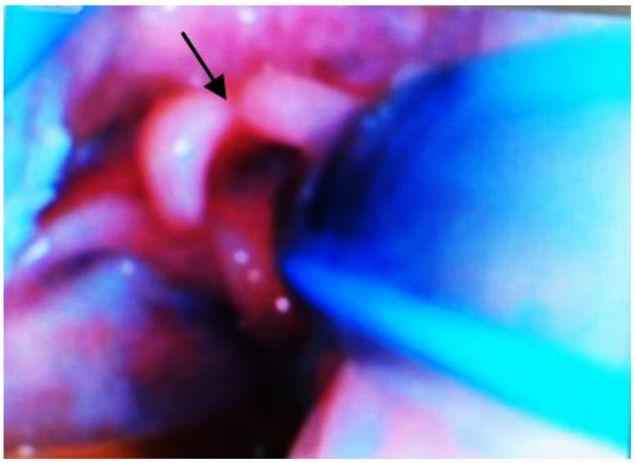

Figure 1: Bifid epiglottis on video-laryngoscope with ETT in situ

The next attempt was taken using a video laryngoscope, blade size 0. With the help of external manipulation, glottic opening was visualised, revealing the presence of a bifid epiglottis (**Figure 1**). ETT size 3mm internal diameter was introduced and fixed at 9 cm after confirming bilateral air entry. The patient was ventilated with 100% oxygen through all the attempts and maintained saturation > 95%. Guided by the video laryngoscope, a size 7 Ryles tube was introduced for gastric decompression. Intraoperatively, the patient remained hemodynamically stable. The procedure was completed uneventfully within an hour. The neuromuscular blockade was reversed with 15 micrograms of neostigmine and 30 micrograms of glycopyrrolate. Tracheal extubation was smooth, and the patient was transferred to the post-operative care unit for monitoring. The patient was discharged one-week post-procedure and is under continuous follow-up for further diagnosis.

Bifid epiglottis, as in our case, is often associated with difficult tracheal intubation as the two halves of the BE often fail to be suspended by the laryngoscope blade, thus obscuring the glottic opening. There may even be symptoms of upper airway obstruction or stridor, under anaesthesia, due to an imbalance of anatomical variations and neural mechanisms favouring airway collapse and increased turbulent airflow.

Bifid epiglottis, being an extremely rare occurrence, has been documented rather infrequently in the literature. Larissa Cronje in 2017, reported a case involving a patient with Bardet-Biedl syndrome scheduled for bilateral club foot repair. After induction, the patient developed stridor, initially assumed to be laryngospasm due to light anaesthesia. However, when CPAP manoeuvres proved ineffective, 10 mg of propofol was administered, and a supraglottic device (SGD) was inserted.[5] Post-surgery, direct laryngoscopy revealed a bifid epiglottis, which folded over the larynx and prolapsed over the laryngeal inlet during inspiration, explaining the stridor. The patient was then awakened without complications. Masanori Tsukamoto in 2018, has documented a case of Pallister Hall Syndrome scheduled for dental treatment under General Anaesthesia.[4] Despite a bifid epiglottis, difficult airway was not encountered. However, emphasis has been laid on importance of preparation for difficult airway.

Management of unanticipated difficult airway is definitely a daunting task. More so, hypoxia associated with difficult airway can result in bradycardia and cardiopulmonary arrest in neonates and infants. This is due to the predominant parasympathetic innervation of heart, making these patients more vulnerable to bradycardia in response to hypoxia and noxious stimuli. However, this situation can be managed by following the difficult airway guidelines. All India Difficult Airway Association (AIDAA) has laid down a structured stepwise approach to manage such emergencies (**Figure 2**). It is extremely important to emphasize that the presence of an experienced anaesthetist, use of adjuncts to conventional laryngoscopy, use of supraglottic airway device, mask ventilation between attempts and use of nasal prongs for para oxygenation, are of paramount importance in effectively handling the situation to avoid any untoward consequences.

This case emphasizes the importance of recognizing laryngeal anomalies that may remain undiagnosed until a patient is under general anaesthesia. In situations of unanticipated difficult airway, it is crucial to understand that children’s unique physiology can lead to rapid hypoxemia and bradycardia, which can result in life-threatening consequences. The effective management of such airway emergencies relies on teamwork and strict adherence to established airway protocols, ultimately improving patient outcomes.

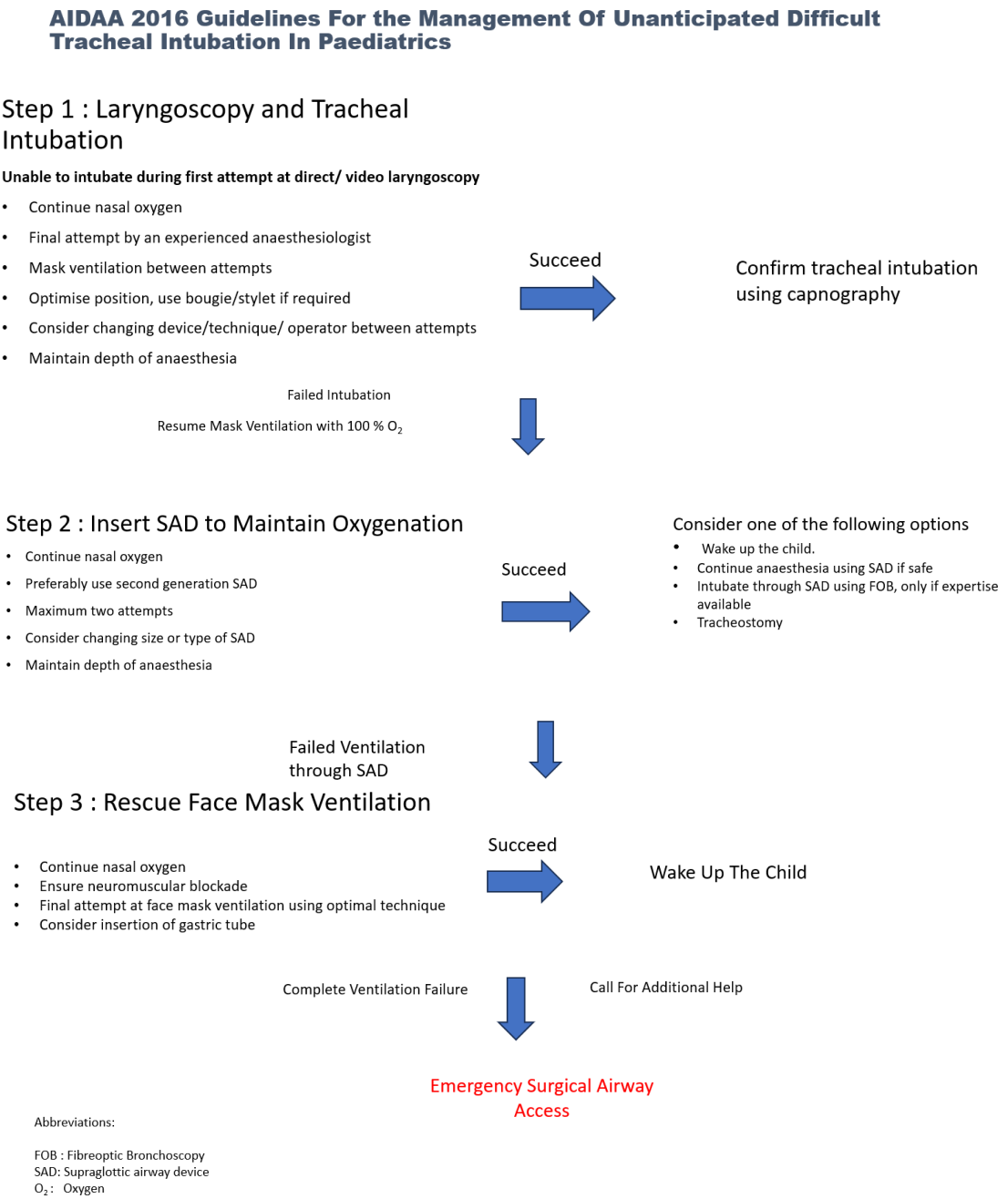

Figure 2: Flow chart based on All India Difficult Airway Association 2016 guidelines for the management of unanticipated difficult tracheal intubation in Paediatrics

1. Conflict of Interest

None.

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