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Combined Fiberoptic and video Laryngoscopic intubation for difficult airway management in a child with turner syndrome and isolated secondary cleft palate

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Abstract
Turner syndrome and craniofacial abnormalities, such as cleft palate, can contribute to significant challenges in airway management due to associated dysmorphic features. Here is the case of a 14 year old girl suspected to have TS with palatal discontinuity scheduled for primary palate repair under general anaesthesia. During the pre-anaesthetic evaluation, she exhibited short stature, a webbed neck, facial dysmorphic features, and restricted mouth opening, indicating a potentially difficult airway. Awake fiberoptic intubation was planned with alternative strategies and airway optimization using local anaesthetics. However, multiple attempts were unsuccessful by a large, edematous epiglottis obscuring the glottic opening. A combined approach utilizing awake fiberoptic bronchoscopy and C-Mac video laryngoscopy successfully facilitated intubation without complications. This case underscores the importance of thorough preoperative assessment, meticulous planning for handling a difficult airway, and the necessity of backup plans to address potential challenges effectively.

Keywords: Turner syndrome, Cleft Palate, Awake Fiberoptic intubation, C-Mac videolaryngoscopy, Difficult Airway, Airway blocks

Introduction
Turner Syndrome (TS) arises from the complete or partial loss of the second X chromosome in phenotypic females, with an occurrence rate of approximately 1 in 2,000 to 3,000 live-born girls. Around half of TS cases exhibit mosaicism, meaning some cells have a 45,X karyotype while others contain different chromosomal arrangements, such as 45,X/46,XX or 45, X/47, XXX. Patients with TS typically exhibit short stature, delayed puberty or ovarian insufficiency, congenital heart and kidney defects, sensorineural hearing loss, vision impairments, thyroid dysfunction, metabolic syndrome, inflammatory bowel disease, and cognitive difficulties [1]. Cleft lip and palate rank among the most prevalent craniofacial anomalies, with isolated cleft palate affecting roughly 1 in 2,000 live births. This condition results from the failure of the nasal and maxillary processes to fuse with the palatine shelves during the eighth week of embryonic development. More than 150 syndromes have been linked with cleft anomalies [2]. The combination of Turner syndrome and cleft palate presents unique challenges during general anaesthesia preparation for difficult airway particularly in airway management.
We present a case of a 14 year old female suspected to have turner syndrome with isolated secondary cleft palate planned for palate repair under general anaesthesia.

Case report
A 14-year-old female, weighing 37 kgs, was admitted with congenital isolated secondary cleft palate and persistent speech difficulties since birth. She was scheduled for primary palatoplasty under general anaesthesia. Pre-operative assessment after obtaining informed parental consent revealed short stature, overriding toes, knock knees, webbed neck with restricted mobility, Elfin facies, retrognathia, inadequate mouth opening (two-finger breadth), Mallampati grade I, and a reduced thyromental distance along with a cleft of the secondary palate. There was a history of second-degree consanguinity. Menstrual history was normal.

MRI of the spine showed mild C4-C5 and C5-C6 disc bulges without compression or stenosis. Cardiac evaluation, routine hematological investigations and chest radiography were normal. In view of challenging airway anatomy, awake orotracheal fiberoptic intubation was planned. A difficult airway cart was prepared, and the patient fasted for eight hours preoperatively. On the morning of surgery, a 20G large bore IV cannula was inserted, and Ringer's Lactate was initiated. IV glycopyrrolate (0.005 mg/kg) was given to reduce secretions. To optimize her airway, she was asked to gargle with 2% lignocaine viscous for 2 minutes, and then nebulized with 2mL of 4% lignocaine. After moving to the operating room, routine monitoring was initiated and vitals recorded. Pre-induction prophylaxis included intravenous ondansetron (0.1 mg/kg) and ranitidine (1.5 mg/kg). Two puffs of Lignocaine 10% spray (each puff delivers 0.1ml, containing 10mg lignocaine) was instilled on the tonsillar pillars and posterior pharyngeal wall. Bilateral superior laryngeal nerve was given with 1.5 ml of 2% lignocaine. Recurrent laryngeal nerve block was given with trans-tracheal instillation of 2% lignocaine. Oxygen was delivered at 2L/min via a nasal cannula throughout induction. Adequate oropharyngeal anaesthesia was confirmed by the patient's tolerance to the insertion of a modified Guedel airway, designed with a longitudinal slit for easy removal after glottic visualization. An initial attempt at awake orotracheal fiberoptic bronchoscopy using a 6.0 mm ID flexometallic endotracheal tube through a modified Guedel airway was unsuccessful due to significantly enlarged, edematous epiglottis and inability to maneuver the scope into the barely visible glottic opening. A second attempt, utilizing a combined approach of awake orotracheal fiberoptic bronchoscopy and simultaneous video laryngoscopy, successfully facilitated intubation with a 6.0 mm ID flexometallic ET tube. Adequate ventilation was confirmed via capnography. Induction was carried out with fentanyl (2 µg/kg), propofol (2 mg/kg), and atracurium (0.5 mg/kg) and maintained with oxygen, nitrous oxide, sevoflurane, and intermittent boluses of atracurium. Intraoperatively, patient was supplemented with inj. paracetamol (15 mg/kg), steroids and tranexamic acid. After completion of surgery and hemostasis confirmation, neuromuscular blockade was reversed using IV glycopyrrolate (0.01 mg/kg) and IV neostigmine (0.05 mg/kg). Following thorough oropharyngeal suctioning, extubation was done once the patient was fully awake, responsive to commands, and generating adequate tidal volume. Post-extubation vitals were stable, and the patient maintained SpO_2 of 99% on room air. She was moved to the recovery area and closely monitored for respiratory distress and aspiration risk.

Discussion

Turner Syndrome (TS) is a complex genetic disorder affecting females, often leading to anatomical and physiological abnormalities, particularly in the airway and cardiovascular system [1]. Cleft palate is commonly associated with structural anomalies such as a high-arched oropharynx, retrognathia, and glossoptosis, which complicate airway management [2]. Thorough preoperative evaluation is crucial to anticipate anaesthetic challenges. In this case, the patient presented with multiple dysmorphic features, including short stature, retrognathia, limited mouth opening, a webbed neck, and restricted cervical mobility, all of which indicated a high likelihood of difficult intubation. Mild cervical spine abnormalities on MRI further necessitated careful head positioning and airway manipulation to prevent neurological complications. Airway Management Plan included:

Awake orotracheal fiberoptic intubation using:

- Modified Guedel's airway [3, 4]
- Video laryngoscope guidance [5-7]

Supraglottic airway device aided endotracheal intubation [8]

Each approach has distinct advantages and limitations. The initial decision to attempt awake orotracheal fiberoptic intubation (AFI) was based on the predicted difficult airway. It is considered the safest approach for managing anticipated difficult airways, but patient cooperation is essential [9, 10]. Adequate airway anaesthesia enhances tolerance to airway devices and suppresses reflexes, improving procedural success. The total dose of lignocaine was maintained within the safe limit of 5 mg/kg. When administered via nebulization and gargling, up to 50% of the lignocaine solution may be lost to the environment, while topical absorption can reach approximately 30% [11]. The World Federation of Societies of Anaesthesiologists (2010) recommended a maximum lignocaine dose of 9 mg/kg, based on lean body weight, for topical anesthesia in adults [12]. To improve the effectiveness and practicality of fiberoptic intubation, various supplementary techniques are frequently employed together. A video laryngoscope serves as a useful aid by providing a clear view of the laryngeal structures and enabling real-time tracking of the fiberoptic bronchoscope's movement [6]. The first attempt was unsuccessful due to challenges in maneuvering the bronchoscope into the obscured glottic opening. Adjusting the technique, a combination of orotracheal fiberoptic bronchoscopy and C-MAC video laryngoscopy proved most effective, emphasizing the value of multimodal airway techniques in managing complex cases [6, 7]. A major contributing factor to the difficult intubation was the large, edematous epiglottis, which significantly obscured the glottic view. Preoperative airway optimization with lignocaine nebulization, nerve blocks, and topical anaesthesia played a significant role in maintaining patient cooperation and minimizing airway reflexes and aiding intubation attempts. A study by Maghawry KM *et al*. evaluating the use of fiberoptic bronchoscopy with or without the C-MAC device in patients with anticipated difficult airways demonstrated that a combined approach significantly shortens intubation time, decreases the duration of a successful attempt, reduces the number of intubation attempts, and improves laryngeal visualization as assessed by Cormack-Lehane grading [5]. The use of intraoperative steroids helped mitigate potential airway edema exacerbation due to multiple intubation attempts.

Conclusion

This patient with Turner's syndrome for cleft palate repair was successfully managed by awake fibreoptic orotracheal intubation assisted with C-mac video laryngoscope without any untoward complications. This case highlights the importance of detailed preoperative assessment and planning for difficult airway management, adequate airway anaesthetization and having alternative strategies and backup plans in place in view of potential complications.

Fig 1: A 14 year old girl with webbed neck, retrognathia and dysmorphic fascies

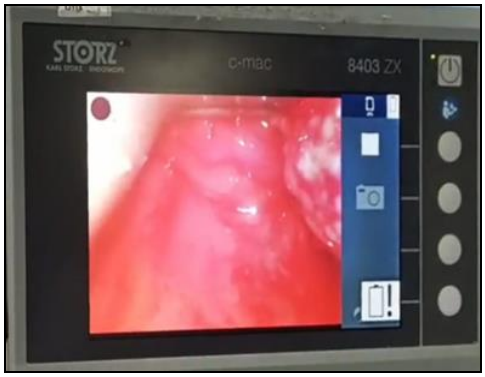

Fig 2: Edematous airway with minimal glottic view on C-Mac videolaryngoscopy

Fig 3: Combined Fibreoptic and video laryngoscopic intubation

Fig 4: Combined Fibreoptic and video laryngoscope aided intubation

Conflict of Interest: Not available

Financial Support: Not available

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