

CASE REPORT

Unexpected failure of videolaryngoscopy due to lingual tonsillar hypertrophy in a patient with Crouzon syndrome and ankylosing spondylitis: a case report

Sung-Mi Ji¹ , Jaegyok Song^{1,*} , Min A. Kwon¹ , Taewon Heo¹, Chang Hoon Seong¹ 

¹Department of Anesthesiology and Pain Medicine, Dankook University College of Medicine, 31116 Cheonan, Republic of Korea

***Correspondence**

12080508@dankook.ac.kr
(Jaegyok Song)

Abstract

Background: Crouzon syndrome and ankylosing spondylitis are independently associated with difficult airway management because of craniofacial abnormalities and severe restriction of cervical spine mobility. Lingual tonsillar hypertrophy is an underrecognized supraglottic pathology that may cause unexpected failure of tracheal intubation. **Case:** A 54-year-old man with Crouzon syndrome and ankylosing spondylitis underwent thoracic spinal surgery. Preoperative airway assessment revealed markedly limited neck extension and a Mallampati class III airway; videolaryngoscopy was selected for tracheal intubation. However, no laryngeal structures were visualized (Cormack-Lehane grade IV), and further attempts were abandoned. After reversal of neuromuscular blockade with sugammadex, the patient was awakened and underwent awake fiberoptic intubation. Fiberoptic examination revealed severe lingual tonsillar hypertrophy with atypical supraglottic soft tissue structures obscuring the epiglottis and laryngeal inlet. Despite technical difficulties with airway blocks, incremental topical anesthesia enabled successful awake fiberoptic intubation while maintaining spontaneous ventilation. The surgery and extubation over an airway exchange catheter were completed uneventfully. **Conclusion:** Videolaryngoscopy may fail in patients with combined craniofacial abnormalities and severe cervical spine rigidity, particularly in the presence of occult supraglottic pathology such as lingual tonsillar hypertrophy. Early recognition of failed intubation, timely transition to awake fiberoptic intubation, and careful extubation planning are essential for safe airway management.

Keywords

Airway management; Ankylosing spondylitis; Crouzon syndrome; Difficult airway; Fiberoptic intubation; Lingual tonsil hypertrophy; Laryngoscopy

1. Introduction

Crouzon syndrome is a craniosynostosis disorder associated with craniofacial dysmorphism that can complicate facemask ventilation and tracheal intubation, making airway management a major anesthetic concern for anesthesiologists caring for affected patients [1, 2]. Ankylosing spondylitis further increases airway risk by causing severe restriction of cervical spine mobility, limiting optimal alignment for direct or videolaryngoscopy and strengthening the rationale for awake techniques that preserve spontaneous ventilation [3]. In addition, lingual tonsillar hypertrophy (LTH) is an underrecognized supraglottic pathology that may cause unexpectedly difficult or failed intubation, despite apparently acceptable preoperative airway findings [4, 5].

Current difficult airway guidelines emphasize early recognition of difficulty, avoidance of repeated attempts, maintenance of oxygenation, and prompt transition to an alternative airway

strategy, including awakening the patient when appropriate [6, 7]. Extubation planning is also critical in patients with difficult intubation, and staged extubation over an airway exchange catheter may reduce the risk when reintubation is anticipated to be challenging [8].

We report a case in which videolaryngoscopy failed in a patient with Crouzon syndrome and ankylosing spondylitis, and awake fiberoptic intubation was successfully performed after identification of LTH and atypical perilaryngeal anatomy.

2. Case presentation

A 54-year-old man (American Society of Anesthesiologists physical status II; weight 65.9 kg, height 168 cm; nonsmoker) with Crouzon syndrome and ankylosing spondylitis was scheduled for a thoracic laminectomy and decompression for progressive motor weakness of both lower limbs. The patient had a history of hypertension. Pulmonary function testing and

echocardiography results were within normal limits. He had previously undergone two-jaw surgery for Crouzon syndrome 25 years earlier.

The patient denied symptoms suggestive of obstructive sleep apnea, dysphagia, voice change, or chronic airway obstruction. Preoperative airway examination revealed a mouth opening of 5 cm, markedly limited neck extension, and a Mallampati class III airway. Awake fiberoptic intubation was considered preoperatively because of cervical spine rigidity; however, videolaryngoscopy was initially selected because the preserved mouth opening and history of previous corrective maxillofacial surgery suggested that adequate glottic visualization would be achievable. The patient's prior uneventful airway management during previous procedures may also have influenced this initial decision-making process.

Upon arrival in the operating room, the team instituted standard monitoring, including electrocardiography, pulse oximetry, invasive arterial blood pressure monitoring through a radial artery catheter, central venous pressure monitoring through a pre-existing right internal jugular venous catheter, and bispectral index monitoring. Initial vital signs were as follows: blood pressure 131/75 mmHg, heart rate 87 beats/min, and respiratory rate 23 breaths/min.

General anesthesia was induced at 10:50 with intravenous lidocaine 1 mg/kg, propofol 1.5 mg/kg, and rocuronium 0.6 mg/kg, and maintained with desflurane at 7 vol%. Mask ventilation was achieved adequately with the aid of an oral airway adjunct. Orotracheal intubation was attempted using a videolaryngoscope (McGRATH™ MAC, Medtronic, Minneapolis, MN, USA) at 10:55; however, no laryngeal structures were visualized (Cormack–Lehane grade IV), and the larynx appeared fixed and immobile despite optimization maneuvers. Facemask ventilation remained adequate using a two-handed technique and an oral airway adjunct. Further videolaryngoscopic attempts were abandoned, and the patient was awakened to facilitate awake fiberoptic intubation. Desflurane was discontinued, and ventilation with 100% oxygen was initiated at 11:10. Neuromuscular blockade was reversed with intravenous sugammadex (200 mg). Following sugammadex administration, the train-of-four count increased from 1/4 to 4/4, spontaneous ventilation resumed, and the patient regained full consciousness with purposeful responses at 11:16. The situation was explained to the patient, and informed consent for awake intubation was obtained.

Dexamethasone (5 mg) and glycopyrrolate (0.2 mg) were administered intravenously. Awake oral fiberoptic intubation was performed in the semi-sitting position while maintaining spontaneous ventilation. No systemic sedation was administered. Supplemental oxygen was continuously delivered via a nasal cannula at 8 L/min throughout the procedure. Topical anesthesia of the oral cavity was achieved using 10% lidocaine spray. Transtracheal injection and superior laryngeal nerve block were unsuccessful because the anatomical landmarks of the anterior neck were difficult to identify, and the anterior neck structures were extremely rigid and fixed, precluding needle penetration and successful completion of the airway blocks. Therefore, topical anesthesia was administered incrementally while maintaining spontaneous ventilation.

A single-use flexible bronchoscope (outer diameter 4.9 mm;

Vathin Medical Instrument Co., Ltd., Xiangtan, Hunan, China) was used to perform awake fiberoptic intubation. During fiberoptic examination, marked lingual tonsillar hypertrophy overlying and encroaching on the epiglottis (Fig. 1) and atypical perilaryngeal soft tissue structures intermittently covering the laryngeal inlet were observed (Fig. 2). A relatively normal-appearing larynx was visualized behind the overlying supraglottic soft tissue structures immediately before tracheal intubation (Fig. 3). Visualization of the vocal cords was initially limited because of these anatomical abnormalities and transient coughing related to incomplete topical anesthesia. Additional topical anesthesia was therefore administered by spraying 2% lidocaine onto the larynx through the working channel of the bronchoscope on three occasions (2 mL each; total dose, 120 mg). Tracheal intubation was subsequently achieved using a 6.5-mm endotracheal tube on the second fiberoptic attempt after additional topical anesthesia.

Awake fiberoptic intubation was initiated at 11:30 and completed at 11:45. The lowest oxygen saturation during the procedure was 97%. Despite transient coughing caused by incomplete topical anesthesia, the patient remained cooperative and tolerated the procedure well.

Following successful awake fiberoptic intubation, anesthesia was re-induced with intravenous propofol and maintained with desflurane and remifentanyl. Additional rocuronium was administered after adequate recovery from the initial neuromuscular blockade had been clinically confirmed. No clinically significant delay in the onset of neuromuscular blockade was observed. The patient was positioned carefully while maintaining neutral alignment of the cervical spine throughout the procedure. A T10–11 laminectomy was then performed over a period of 4 h and 5 min.

3. Discussion

This case demonstrates how multiple independent mechanisms can converge to result in complete videolaryngoscopy failure. Importantly, although this patient was recognized preoperatively as having a potentially difficult airway because of Crouzon syndrome and ankylosing spondylitis, the severity and underlying mechanism of intubation failure were not fully predictable before airway instrumentation.

Crouzon syndrome is associated with craniofacial and upper airway anatomical variations that may impair visualization of the glottis, even when mouth opening appears adequate [1, 2]. In this case, the patient had previously undergone corrective maxillofacial surgery and demonstrated preserved mouth opening. These findings, together with prior uneventful airway management in similar patients, may have contributed to the initial selection of videolaryngoscopy. However, this case highlights the limitations of relying on apparently reassuring airway findings or prior uneventful clinical experience when multiple predictors of difficult airway are present. In retrospect, the coexistence of Crouzon syndrome, severe cervical immobility due to ankylosing spondylitis, Mallampati class III airway, and occult supraglottic pathology may have warranted stronger consideration of awake fiberoptic intubation as the primary airway strategy.

In the present case, occult lingual tonsillar hypertrophy and

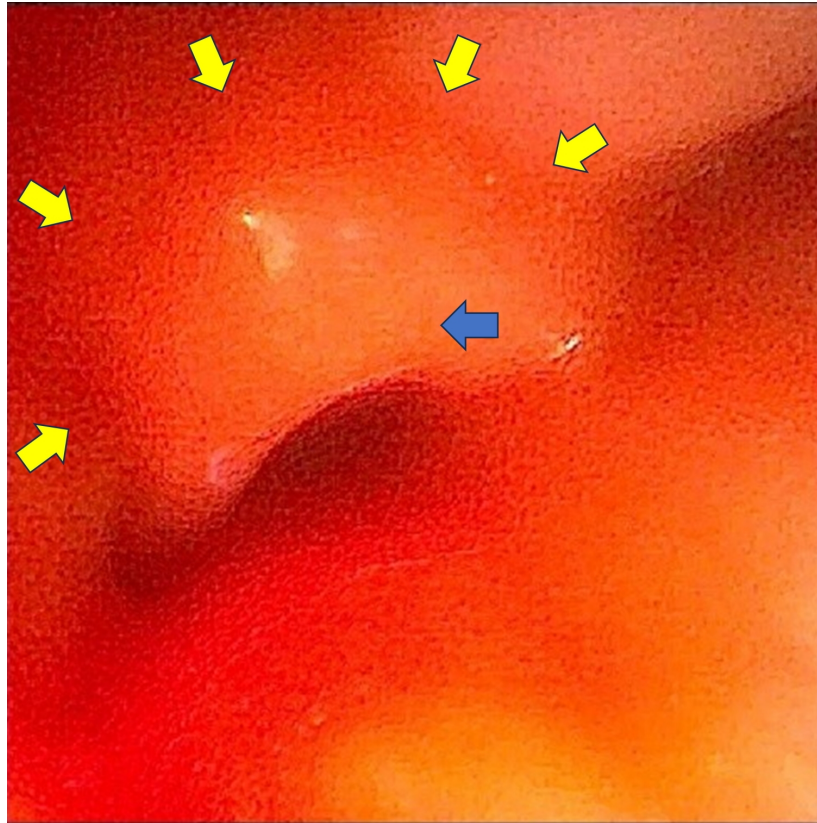


FIGURE 1. Marked lingual tonsillar hypertrophy (yellow arrows), surrounding and partially obscuring the epiglottis (blue arrow), resulting in near-complete obscuration of the laryngeal inlet.

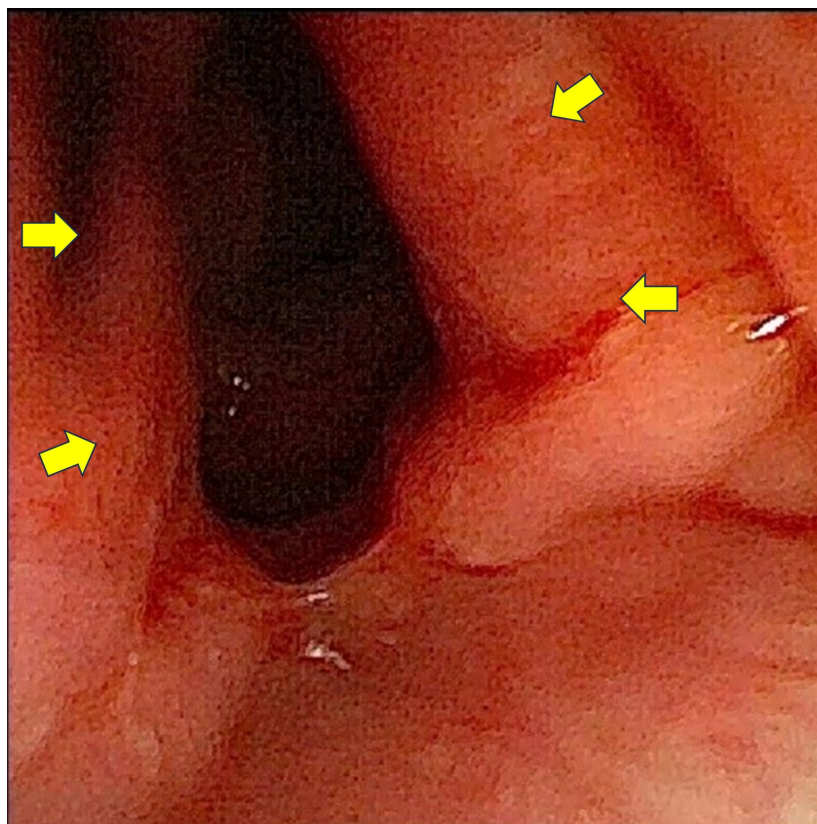


FIGURE 2. A curtain-like, mobile perilaryngeal soft tissue structure (yellow arrows) intermittently obscuring the epiglottis and laryngeal inlet.

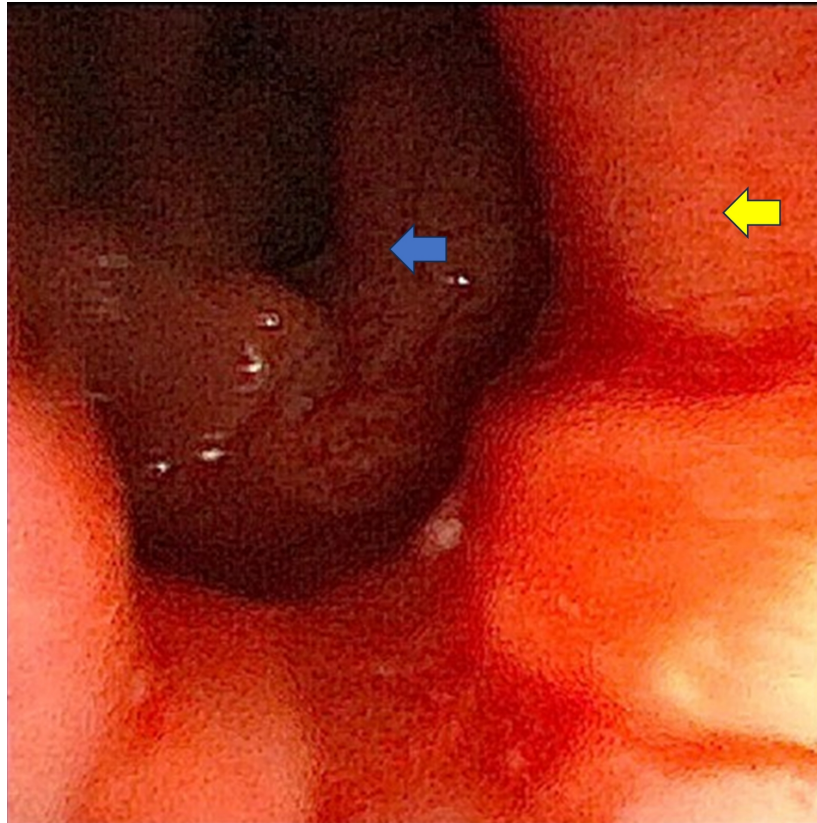


FIGURE 3. Laryngeal view just before endotracheal intubation. A relatively normal-appearing larynx (blue arrow) is visualized behind the overlying supraglottic soft tissue structures (yellow arrow).

severe cervical immobility likely contributed to the unexpectedly severe difficulty with airway management.

The most decisive finding during awake fiberoptic intubation was marked lingual tonsillar hypertrophy, with epiglottic encroachment and atypical perilaryngeal structures intermittently covering the laryngeal inlet. Lingual tonsillar hypertrophy is a well-described but often overlooked cause of unexpectedly difficult airways and may not be detected during routine preoperative airway assessments [4, 5]. In this case, the marked lingual tonsillar hypertrophy appeared to be a major anatomical contributor to the failed videolaryngoscopy. The enlarged lingual tonsils appeared to obscure the vallecular space and may have impaired effective elevation of the epiglottis, resulting in complete failure of glottic visualization despite optimization maneuvers. Collectively, these anatomical abnormalities and severe clinical immobility created a high-risk airway scenario in which videolaryngoscopy yielded a Cormack–Lehane grade IV view.

During subsequent awake fiberoptic examination, dynamic movement of the supraglottic soft tissue was additionally observed and may have further complicated airway instrumentation. This case also illustrates that an anticipated difficult airway does not necessarily predict the precise mechanism or severity of failed intubation.

Importantly, this case highlights the limitations of external airway features and standard bedside airway assessments in predicting severe supraglottic pathology. Even when conventional predictors suggest that airway management may be feasible, clinically silent supraglottic abnormalities may result

in complete videolaryngoscopy failure, underscoring the need for early reassessment.

A major strength of this case was the decision to abandon further videolaryngoscopic attempts and awaken the patient to facilitate awake fiberoptic intubation. This approach aligned with current American Society of Anesthesiologists and Difficult Airway Society guidelines, which emphasize limiting repeated airway instrumentation, reassessing the airway strategy when initial attempts fail, and prioritization of oxygenation [6, 7].

Awake fiberoptic intubation allowed maintenance of spontaneous ventilation while minimizing cervical spine movement, consistent with contemporary recommendations and reports in patients with severe ankylosing spondylitis [3]. As airway blocks were technically challenging because of the marked rigidity of the anterior neck structures, incremental administration of topical anesthesia via the working channel of the bronchoscope facilitated successful intubation. Extubation was appropriately managed as a high-risk phase of airway management, and staged extubation over an airway exchange catheter provided an additional margin of safety, consistent with contemporary recommendations [8].

4. Conclusion

In patients with combined Crouzon syndrome and ankylosing spondylitis, occult lingual tonsillar hypertrophy may contribute to severe or complete videolaryngoscopy failure despite apparently reassuring preoperative airway findings. This case

emphasizes the importance of promptly abandoning unsuccessful videolaryngoscopic attempts, transitioning to awake fiberoptic intubation with maintenance of spontaneous ventilation, and planning staged extubation when reintubation is anticipated.

ABBREVIATIONS

LTH, lingual tonsillar hypertrophy.

AVAILABILITY OF DATA AND MATERIALS

All data generated or analyzed in this study are included in the published article. Additional information is available from the corresponding author upon request.

AUTHOR CONTRIBUTIONS

SMJ and TH—collected and analyzed the clinical data. CHS, JS and MAK—drafted the manuscript. All authors critically revised the manuscript and approved the final version.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Institutional review board approval was obtained from Dankook University Hospital (DKUH 2026-01-001). Written informed consent was obtained from the patient for publication of this case report and the accompanying images.

ACKNOWLEDGMENT

The authors gratefully acknowledge the institutional support of Dankook University Hospital.

FUNDING

This research received no external funding.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

REFERENCES

- [1] Fernandes M, Eufrazio A, Bonifacio J, Marcelino J. Airway management in a patient with Crouzon syndrome proposed to orthognathic surgery. *BMJ Case Reports*. 2018; 2018: bcr2017219371.
- [2] OrphanAnesthesia. Crouzon syndrome: anaesthesia recommendations. 2026. Available at: <https://www.orphananesthesia.eu/en/rare-diseases/published-guidelines/crouzon-syndrome.html> (Accessed: 27 May 2026).
- [3] Rebai L, Kalai F, Ardhaoui I, Brahem SB, Rabhi B, Fakhfakh H. Approaches to difficult airway management in a patient with ankylosing spondylitis and severe cervical spine deformities. *International Journal of Surgery Case Reports*. 2025; 130: 111260.
- [4] Kwon MA, Song J, Park K. Difficult airway management in a case with lingual tonsil hypertrophy and temporo-mandibular joint partial ankylosis. *Korean Journal of Anesthesiology*. 2013; 65: 359–360.
- [5] Wong P, Sleight JW. Airway management of lingual tonsillar hypertrophy: a narrative review. *Anaesthesia and Intensive Care*. 2024; 52: 16–27.
- [6] Apfelbaum JL, Hagberg CA, Connis RT, Abdelmalak BB, Agarkar M, Dutton RP, *et al*. 2022 American Society of Anesthesiologists practice guidelines for management of the difficult airway. *Anesthesiology*. 2022; 136: 31–81.
- [7] Ahmad I, El-Boghdady K, Iliff H, Dua G, Higgs A, Huntington M, *et al*. Difficult Airway Society 2025 guidelines for management of unanticipated difficult tracheal intubation in adults. *British Journal of Anaesthesia*. 2026; 136: 283–307.
- [8] Heisenberg D, Stieger A, Oberle F, Parotto M, Heidegger T. Challenge of tracheal extubation and tube exchange of a difficult airway: a narrative review. *Journal of Anesthesia*. 2025; 39: 465–475.

How to cite this article: Ji SM, Song J, Kwon MA, Heo T, Seong CH. Unexpected failure of videolaryngoscopy due to lingual tonsillar hypertrophy in a patient with Crouzon syndrome and ankylosing spondylitis: a case report. *Signa Vitae*. 2026. doi: 10.22514/sv.2026.076.