

Case Report

Complicated Laryngotracheal Stenosis Occurring Early after COVID-19 Intubation

Kirsten Wong ¹, Alden Smith ², Jose Alonso ^{3,4} and Jennifer Long ^{1,3,4,*}¹ David Geffen School of Medicine, University of California, Los Angeles, CA 90095, USA² Department of Otolaryngology—Head & Neck Surgery, Stanford University School of Medicine, Palo Alto, CA 94304, USA³ Department of Head and Neck Surgery, David Geffen School of Medicine at UCLA, Los Angeles, CA 90095, USA⁴ Greater Los Angeles VA Healthcare System, Los Angeles, CA 90073, USA

* Correspondence: jlong@mednet.ucla.edu; Tel.: +1-310-825-3015

Abstract: Background: Airway stenosis is a known complication of prolonged intubation in hospitalized patients. With the high rate of intubations in patients with COVID-19 pneumonia, laryngotracheal stenosis (LTS) is a complication of COVID-19 that drastically reduces quality of life for patients who may remain tracheostomy-dependent. Methods: Patient medical history, laryngoscopy, and CT imaging were obtained from medical records. Results: We report four cases of complicated LTS following intubation after COVID-19 pneumonia and explore the current literature in a narrative review. Four patients developed LTS following intubation from COVID-19 pneumonia. Three patients remain tracheostomy-dependent, and the fourth required a heroic operative schedule to avoid tracheostomy. Conclusion: Intubation for COVID-19 pneumonia can result in severe LTS, which may persist despite endoscopic intervention.

Keywords: tracheal stenosis; subglottic stenosis; COVID-19; intubation



Citation: Wong, K.; Smith, A.;

Alonso, J.; Long, J. Complicated Laryngotracheal Stenosis Occurring Early after COVID-19 Intubation. *Surgeries* **2022**, *3*, 211–218. <https://doi.org/10.3390/surgeries3030023>

Academic Editor: Cornelis F.M. Sier

Received: 8 July 2022

Accepted: 1 August 2022

Published: 5 August 2022

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1. Introduction

Throughout the COVID-19 pandemic, endotracheal intubation has been a mainstay of treatment for patients with severe disease. Although airway management has shifted from early intubation to prolonged non-invasive ventilatory support [1], intubation rates among patients requiring supplemental oxygen were estimated at 30% [2,3]. Early pandemic mortality reports from China, Italy, and Spain were up to 88% in intubated patients, although prone positioning, corticosteroids, and biologic and antiviral treatments have improved survival [4–6]. The United States has had seven million COVID-19 cases since September 2020, with in-hospital mortality rates of 23–67% in intubated patients [7]. Survivors of COVID-19 intubation of any duration are also at risk for glottic and subglottic stenosis (SGS) [8–16].

Prolonged intubation has long been associated with the development of airway stenosis. However, early case–control series demonstrate higher stenosis incidence in COVID-19 patients when compared to other patients with similar lengths of intubation. Fiacchini et al. followed 30 COVID-19 patients with severe respiratory failure and 45 patients without COVID-19 who underwent invasive mechanical ventilation for more than 14 days. A total of 47% of the COVID-19 patients demonstrated airway complications compared to 2% of the control group [17].

This report introduces four cases of devastating LTS after extubation from COVID-19. We highlight the heterogeneity of presentations and suggest future directions for research in the care of these patients.

2. Materials and Methods

The institutional review board considered this retrospective analysis exempt from review. Patient medical history, laryngoscopy, and CT imaging were obtained from medical records over the past two years since the early beginnings of the pandemic to the current date of publication.

3. Results

3.1. Case Descriptions

3.1.1. Case 1

A 73-year-old edentulous male with a history of COVID-19 pneumonia and 10-day intubation at an outside hospital developed worsening dyspnea 2.5 weeks after extubation. He experienced acute respiratory decline with inspiratory stridor, tracheal retractions, a high-pitched cough, and hypoxia. Flexible laryngoscopy identified grade 3 SGS, 71–99% stenosis by the Cotton–Meyer grading scale [18], with substantial edema, and chest CT identified a hyperdense foreign body in the proximal left mainstem bronchus (Figure 1a).

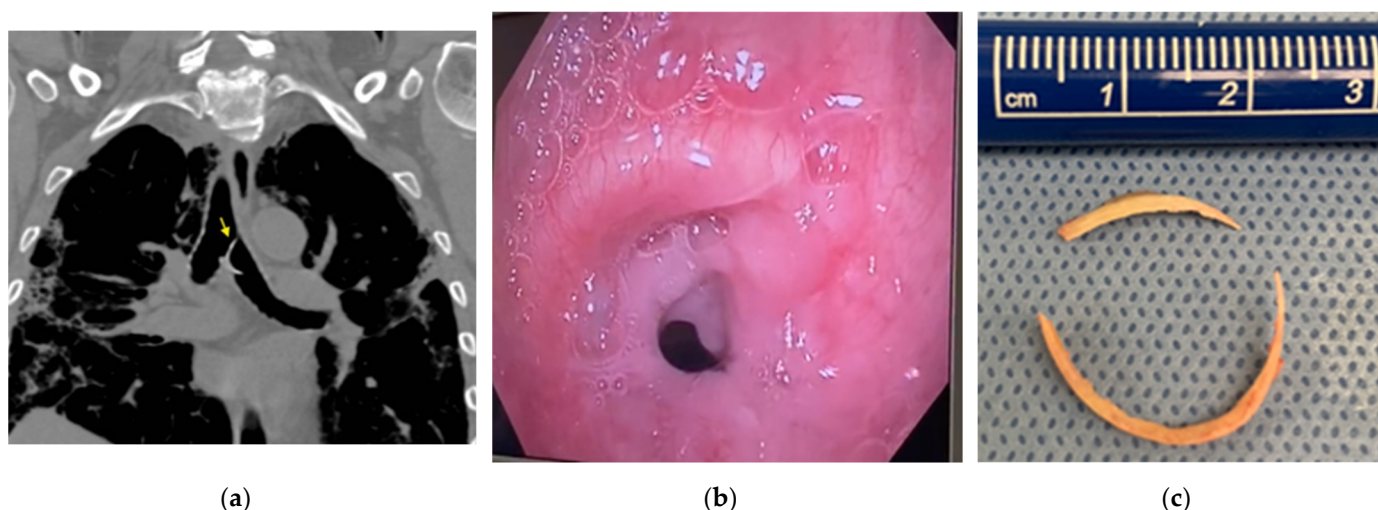


Figure 1. (a) SGS occurring at the cricoid cartilage, which was fractured and is protruding into the airway lumen (yellow arrow); (b) non-contrast chest CT scan, coronal view showing a curvilinear hyperdense foreign body in the proximal left mainstem bronchus marked with arrow; (c) gross appearance of foreign body.

Endoscopic management in the operating room began with flexible bronchoscopy and laryngeal mask anesthesia [19]. This identified a grade 3 SGS at the level of the upper cricoid which the bronchoscope could not pass through (Figure 1b). The stenosis was dilated using a controlled radial expansion balloon inflated to 14 mm through the bronchoscope working channel. Airway caliber significantly improved, and a 5–0 endotracheal tube was inserted without resistance. Subsequent rigid bronchoscopy identified a whitish, jagged, semicircular foreign body in the proximal mainstem bronchus, which was removed with endoscopic grasping forceps (Figure 1c). Final pathology described the foreign body as cartilage with calcifications consistent with cricoid fracture.

After responding well to initial endoscopic management, he was noted on office laryngoscopy two weeks later to have recurrent granulation tissue at the level of the injured cricoid cartilage. He required several endoscopic treatments with laser, dilation, and steroid injection to control the recurrent SGS. He wished to avoid tracheostomy and was treated with an aggressive bronchoscopy schedule every two weeks for a full year. At that time, his tracheal diameter was stable, and his breathing was not impaired, so the surgical interval was increased.

3.1.2. Case 2

A 77-year-old male with sleep apnea and coronary artery disease developed concomitant *Pseudomonas aeruginosa* and COVID-19 pneumonia in spring 2020, requiring intubation for 10 days until tracheostomy. His course was complicated by venous thromboembolism and end-stage renal disease requiring dialysis. The patient's tracheostomy was decannulated three months after recovery, but he later developed progressive dyspnea over the following three months. On laryngoscopy, he was found to have bilateral vocal cord hypomobility, a large subglottic granuloma, and edema of the infraglottis (Figure 2a,b). Anterior cricoid fracture was seen on CT imaging with an anterior shelf visualized endoscopically.

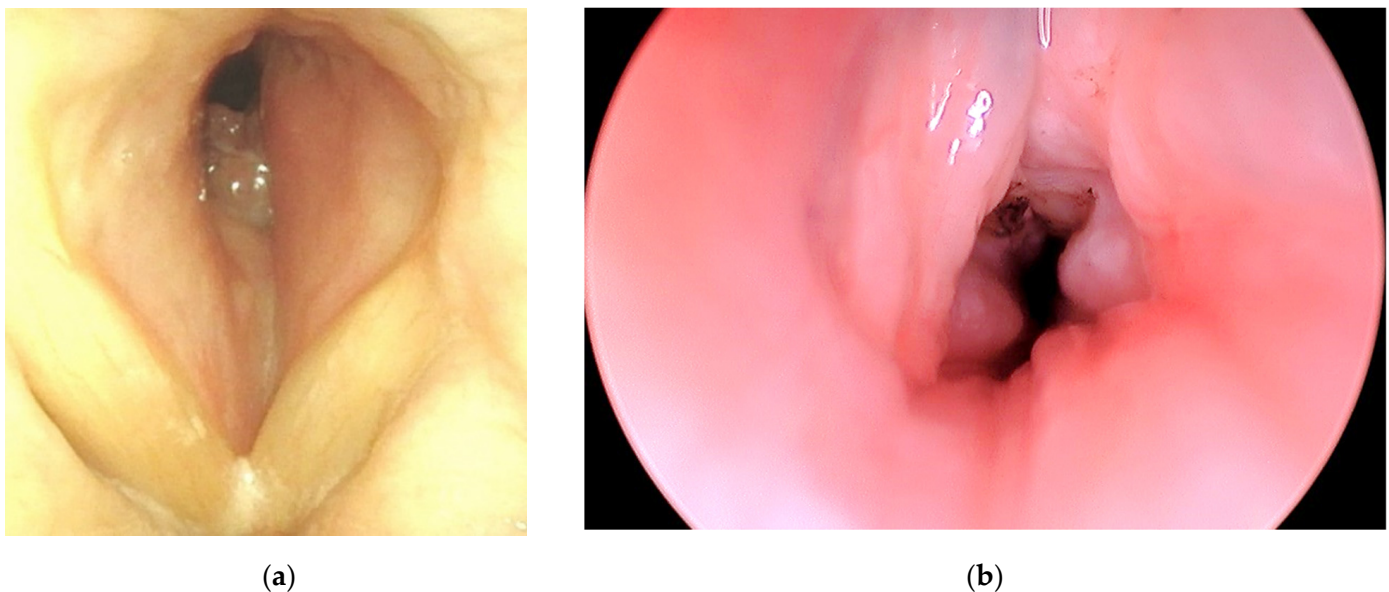


Figure 2. (a) Laryngoscopy at initial presentation demonstrated infraglottic edema and anterior granulation tissue causing airway obstruction; (b) endoscopic images of patient after last endoscopic procedure to treat infraglottic airway narrowing.

Initial management of his airway stenosis included three endoscopic interventions over four weeks to debulk the subglottic granuloma with CO₂ laser excision, dilation of LTS, and triamcinolone depot injection. Despite these efforts, he required tracheostomy replacement to manage respiratory distress from persistent airway narrowing. The patient had three additional endoscopic procedures including CO₂ laser arytenoidectomy and cordotomy and an open transcervical reduction in the fractured anterior cricoid. In the most recent endoscopic intervention, persistent infraglottic edema was treated with CO₂ laser division and balloon dilation. He also exhibited intense laryngeal hyperadduction, which has responded to bilateral botulinum toxin injection at three-month intervals. These measures improved airway patency to allow for voicing, but the patient remains tracheostomy-dependent.

3.1.3. Case 3

An 80-year-old male with congestive heart failure, hyperlipidemia, and hypertension had COVID-19 pneumonia in late 2020. After hospitalization for one month, he was intubated for 12 days, then underwent tracheostomy and was decannulated after six weeks. Two months after decannulation, the patient presented with stridor and bilateral vocal fold immobility (Figure 3a) and was admitted for urgent revision tracheostomy with direct laryngoscopy. He was found to have grade 4 posterior glottic stenosis (PGS) and no detectable lumen according to the Cotton–Meyer grading scale [18], with bilateral cricoarytenoid joint fixation. After CO₂ laser posterior scar division, the patient was discharged home with a tracheostomy tube and tolerated use of a one-way speaking valve.

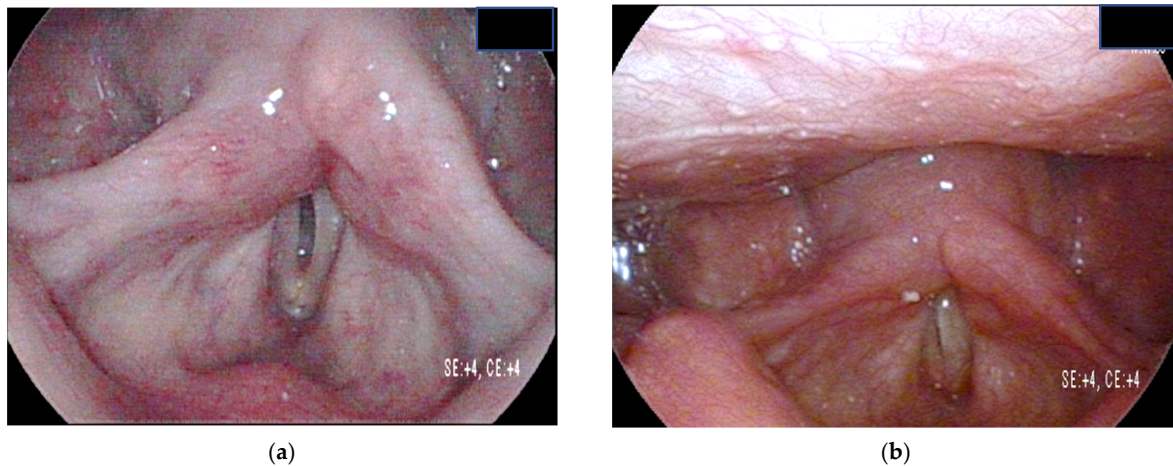


Figure 3. Stroboscopy images of Case 3 (a) preoperatively with maximal abduction without tracheostomy and (b) post-arytenoidectomy and tracheostomy showing increased spasm.

Four months after revision tracheostomy placement, the patient developed progressive difficulty speaking and one-way speaking valve intolerance. Videolaryngostroboscopy (VLS) demonstrated complete fixation of the posterior glottis and laryngeal hyperadduction. He subsequently underwent endoscopic-laser-assisted right arytenoidectomy, suture lateralization of the true vocal fold, and botulinum toxin chemodenervation of the bilateral vocal fold adductors. Following surgery, the patient noted less vocal strain and easier voicing with finger occlusion of the trach, but he remains tracheostomy-dependent (Figure 3b).

3.1.4. Case 4

A 60-year-old male with COVID-19 pneumonia in fall 2020 was intubated for one month. Three months after extubation, the patient presented with urgent airway distress and underwent tracheostomy. Six months later, he remained tracheostomy-dependent and was found to have bilateral vocal fold immobility with no glottic opening on VLS (Figure 4a). Direct laryngoscopy under general anesthesia demonstrated grade 4 PGS with complete fixation of bilateral arytenoid cartilages and extensive laryngeal spasm of the true and false vocal folds. He underwent two endoscopic treatments of CO₂ laser scar division and botulinum toxin injection to bilateral vocal folds to reduce laryngeal spasm. His voicing improved, but VLS showed persistent vocal fold fixation (Figure 4b), for which he remains tracheostomy-dependent and declines further treatment.

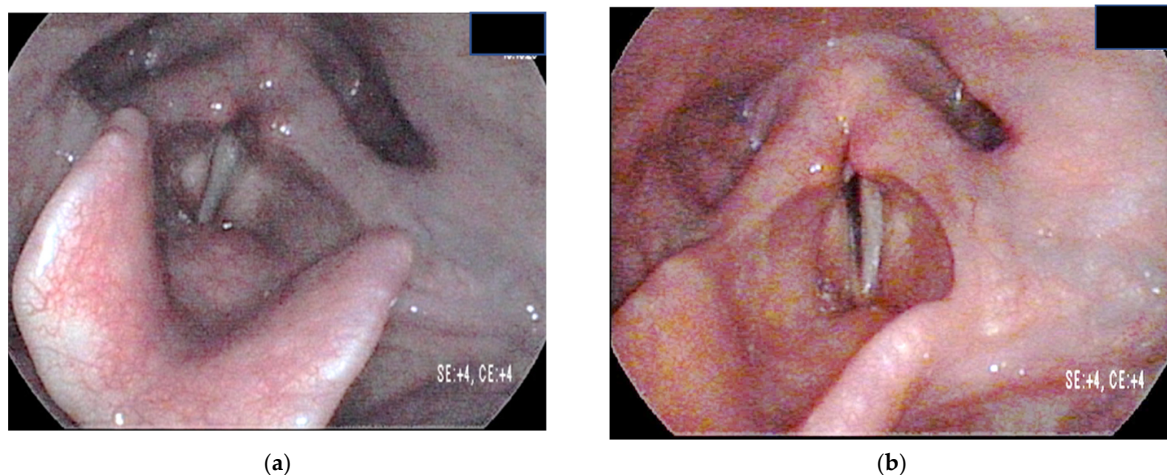


Figure 4. Stroboscopy images of Case 4 (a) preoperatively with maximal abduction and (b) post-arytenoidectomy.

4. Discussion

The massive numbers of intubated patients seen in the COVID-19 pandemic have brought renewed urgency to increasing awareness of intubation complications. There were seven million cases of COVID-19 in the United States by September 2020, with a 30% intubation rate among patients requiring oxygen supplementation [3,7]. Of these patients, extubation rates ranged from 35 to 57% with tracheostomy rates of up to 18% [20–22]. Patients intubated for COVID-19 pneumonia have a higher incidence of airway-related complications when compared to non-COVID patients [17]. While severe airway-related complications are rare, it is anticipated that the high number of intubated COVID-19 survivors may contribute to increased incidence of airway stenosis [5]. This case series highlights the severity of intubation-related complications in these patients.

Airway-related complications with varying degrees of severity have been reported in patients intubated for COVID-19 pneumonia. In a prospective series following 13 patients hospitalized and intubated for COVID-19, all patients had abnormalities on laryngoscopy, including unilateral vocal fold immobility (40%), PGS (15%), and SGS (15%) [23]. Vocal cord palsy and ulcers, laryngotracheal granulomas, malacia, and tracheal necrosis have also been reported [5,24,25].

Prior to the COVID-19 pandemic, rates of post-intubation and post-tracheostomy airway stenosis ranged from 10 to 22%, with prolonged intubation being the primary risk factor [26]. Patients intubated for longer than 10 days had a higher risk for developing PGS, and each additional day of intubation increased the odds by 21% [27]. Throughout the pandemic, the median duration of endotracheal intubation in COVID-19 survivors has remained stable at 17 days [28,29]. Longer durations of intubation for COVID-19 pneumonia may explain higher instances of airway complications. Thus, early tracheostomies have been recommended to avoid prolonged endotracheal intubation in both COVID-19 and non-COVID-19 patients [30,31].

While prolonged intubation is the primary risk factor for developing acute laryngeal injury (ALI) or LTS, mechanical injury and inflammation may also increase risk. Mucosal ulceration has been observed in non-COVID-19 patients intubated for as few as 12 h [27,32,33]. Furthermore, one meta-analysis in non-COVID-19 patients demonstrated an 8% incidence of LTS even in patients with early conversion to tracheostomy [30]. In a cohort of 100 non-COVID-19 patients, ALI was independently associated with mechanical injury from large endotracheal tubes (>7.0) and the pro-inflammatory conditions of obesity and type II diabetes [32]. These conditions are known to be highly prevalent in individuals with severe COVID-19 infections.

When controlled for length of intubation, COVID-19 patients still demonstrate greater incidence and severity of airway complications. Fiacchini et al. compared 30 COVID-19 patients and 45 non-COVID-19 patients who both underwent prolonged mechanical ventilation courses. When evaluated by bronchoscopy for airway complications, 47% of COVID-19 patients demonstrated complications compared to 2% of the control group [17]. It can be hypothesized that rapid LTS in COVID-19 patients may be due to a combination of mucosal damage from large endotracheal tubes and pronation maneuvers, weakened tracheal mucosa from viral replication or high steroid doses, hypoxic damage, and decreased healing from a heightened COVID-19 inflammatory environment [17,34,35]. The relative contributions of these COVID-specific insults versus underlying comorbid medical conditions remain to be defined.

In this series, length of intubation prior to tracheostomy or extubation ranged from 10 days to one month, which may account for the severity of complications we report. Patients' presentations occurred weeks to months after recovery from COVID-19. All patients presented with respiratory distress caused by airway injuries including SGS, PGS, airway granulation tissue, laryngeal spasm, and vocal cord hypomobility. Of note, three of four patients demonstrated bilateral vocal cord hypomobility at presentation, and two benefited from Botox injection for laryngeal hyperadduction. There were also two cases

in this series with cricoid cartilage fracture—a rare airway complication which may be due to intubation-related trauma or a complication of tracheostomy [36].

LTS can be treated with endoscopic or open management [37–39]. Endoscopic management includes balloon dilation, carbon dioxide laser ablation, or cold knife. Adjunctive treatments include mitomycin or glucocorticoid depot injection [38]. For severe stenoses, open techniques of full resection or laryngotracheoplasty facilitate long-term decannulation [39]. Medical comorbidities are known to worsen outcomes of open airway reconstruction in adults, including prior tracheostomy, diabetes, and possibly laryngopharyngeal reflux [40,41]. No patients in this series underwent open airway reconstruction: two were considered poor candidates due to severe medical comorbidities, one was offered staged laryngotracheoplasty and declined, and one was able to remain tracheostomy-free with frequent endoscopic management.

This series demonstrates the severity of COVID-19-related LTS after prolonged intubation. Anecdotally, many practitioners are discussing the long-term airway complications of COVID-19. Despite this widespread experience, representation in the scientific literature is surprisingly sparse, as we have identified in the current literature review. We therefore presented a subset of patients from our institution who illustrate refractory disease. Recognition of this severe complication is important for the broader discourse of COVID-19 complications, as we anticipate an increase in COVID-19 airway-related complications given the prolonged course of the pandemic. Early tracheostomy may be helpful in preventing LTS in COVID-19 patients. Future directions in management may include early identification and intervention [42]. A deeper understanding of the pro-inflammatory environment of COVID-19 infection may also guide our treatment strategies and prevention of airway complications in these patients.

5. Conclusions

LTS is a complication of COVID-19 that may require multiple corrective surgeries, and in severe cases, leave patients tracheostomy-dependent. Tracheal stenosis, while a known complication of prolonged intubation, is anecdotally presenting with greater severity in COVID-19 patients. Through this series of cases, we unequivocally demonstrate the severity and heterogeneity of presentations, which we will continue to observe given the diversity of post-intubation COVID-19 survivors. Mechanisms for an increased risk of LTS are still being studied, and continued attention to COVID-19 survivors will enable early detection of LTS and aid in risk stratification.

Author Contributions: Conceptualization, J.L., A.S., J.A. and K.W.; methodology, J.L., A.S., J.A. and K.W.; formal analysis, A.S. and K.W.; investigation, J.L., A.S., J.A. and K.W.; resources, J.L., A.S. and J.A.; data curation, J.L., A.S. and J.A.; writing, J.A. and K.W.; writing—review and editing, J.L., A.S. and K.W.; visualization, A.S. and K.W.; supervision, J.L.; project administration, J.L.; All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: Not applicable. The institutional review board considered this retrospective study exempt from review.

Informed Consent Statement: Not applicable.

Data Availability Statement: Not applicable.

Acknowledgments: Additional support provided by UCLA's Jonsson Comprehensive Cancer Center (J.L.).

Conflicts of Interest: The authors declare no conflict of interest.

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