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Laryngotracheal Stenosis Following Intubation for COVID-19 Pneumonia: A Report of Two Cases

Derar Al-Domaidat[1], Mohammed Algarni[2], Ro'a Alshaikh Hasan[3], Hosam Amoodi[1,4*]

[1]Otolaryngology Department, Dr. Soliman Fakeeh Hospital, Jeddah, Saudi Arabia
[2]Otolaryngology Department, King Saud Bin Abdulaziz University for Health Science, Jeddah, Saudi Arabia
[3]Internal Medicine Department, Dr. Soliman Fakeeh Hospital, Jeddah, Saudi Arabia
[4]Department of Surgery, Jeddah University, Jeddah, Saudi Arabia
Email: *haamoodi@fakeeh.care

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Abstract

Benign laryngotracheal stenosis (LTS) is a debilitating and potentially life-threatening condition that is commonly caused by iatrogenic events as a result of endotracheal intubation or tracheostomy. Numerous cases are being published for patients with coronavirus disease 2019 (COVID-19) who end up with severe LTS after prolonged intubation or tracheostomy. Here, we presented two cases of LTS due to prolonged intubation after severe COVID-19 pneumonia. The characteristic of these two cases is that both of them needed second time intubation and were readmitted because of severe dyspnoea and all the workup for post-COVID-19 complications were investigated except the LTS which was later diagnosed after one month of suffering of these patients.

Keywords

Subglottic Stenosis, Tracheal Stenosis, Laryngotracheal Stenosis, Prolonged Intubation, COVID-19

1. Introduction

Coronavirus disease 2019 (COVID-19), the viral illness caused by the novel coronavirus severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2), has resulted in significant morbidity and mortality across the world since it erupted in Wuhan China, in December 2019 [1].

Patients who recover from COVID-19 may experience persistence of clinical symptoms beyond four weeks from the onset of acute symptoms. The Center for

Disease Control (CDC) has formulated "post-COVID conditions" to describe health issues that persist more than four weeks after being infected with COVID-19 [1]. The typical clinical symptoms in "long COVID" are tiredness, dyspnea, fatigue, brain fogginess, autonomic dysfunction, headache, persistent loss of smell or taste, cough, depression, low-grade fevers, palpitations, dizziness, muscle pain, and joint pains [1] [2].

COVID-19 was the third leading cause of death in the US in 2020 after heart disease and cancer, with approximately 375,000 death reported. The WHO's current estimate of the global case fatality rate for COVID-19 is 2.2%. However, the case fatality rate is affected by factors that include age, underlying pre-existing conditions, and severity of illness and significantly varies between countries [3].

Several variants of SARS-CoV-2 have been described during the course of this pandemic, among which only a few are considered variants of concern by the WHO, given their impact on global public health [3]. Several vaccines for COVID-19, have been developed. All vaccines appear to be safe and effective tools to prevent severe COVID-19, hospitalization, and death against all variants of concern, but the quality of evidence greatly varies depending on the vaccines considered [4].

COVID-19 may lead to intensive care unit (ICU) admission in up to 12% of all positive cases for massive interstitial pneumonia, with possible long-term endotracheal intubation for mechanical ventilation and subsequent tracheostomy [5].

Post-intubation tracheal stenosis is a rare event, and its incidence is estimated to be 4.9 cases per million per year [4]. However, as a consequence of the combination of an increased number of long-term intubated of critically ill patients, in association with delayed tracheostomy, it is probable that, in the near future, an unprecedented rise of iatrogenic sequelae ranging from granulomas, webs, and LTS, to tracheomalacia, tracheal necrosis, tracheo-esophageal, and tracheo-innominate fistulae may come to arise [6].

In this article, we report two cases of such complication. The unique about them is that both after being improved were readmitted and re-intubated for a second time. Both cases underwent many invasive and non-invasive investigations until finally diagnosed with LTS after much morbidity and cost for them and the health care system. Therefore, in this paper, we want to concentrate on the fact that LTS is a true and serious complication for patients who recover from COVID-19 after invasive mechanical ventilation. This should be taken into consideration for all health care givers who deal with such patients.

2. Case Presentation

Case 1

A 46 year-old female, previously healthy, non-vaccinated for COVID-19, morbidly obese with body mass index (BMI) > 40, came to ER with a one-week history of fever, generalized mayalgia and cough with progressive shortness of breath.

On examination, she was febrile and tachypnic with O_2 saturation of 92% on room air. Chest examination showed bilateral crepitations at the lungs' bases. Laboratory investigations showed high CRP level (173) and normal white blood cell count and differentials. Chest x-ray showed bilateral lung interstitial coarsening and patchy infiltrates. COVID-19 was suspected and nasopharyngeal swab was sent and it came positive after 2 days. The patient was admitted for further supportive treatment.

During admission, the patient's demand for oxygen increased gradually, her general and respiratory condition was getting worse until she started to develop hypoxia and severe respiratory distress for which she was transferred to ICU where she was intubated and connected to a mechanical ventilator. In the ICU, she developed septic shock and acute kidney injury and necessitated hemodialysis.

After 15 days of intubation and giving medical and supportive therapy, she was extubated. Then she improved enough to be transferred to general medical ward. Finally, she was discharged home on O_2 nasal cannula after 3 weeks of admission.

Two weeks following discharge, she presented to ER again with severe hypoxia (O_2 saturation was 30%) with decreased level of consciousness. Therefore, she was intubated again in the ER and admitted to ICU. After admission, she was prone to many investigations and procedures to exclude the long list of differential diagnosis which might explain her condition, such as myocarditis, myocardial infarction, stroke, pulmonary embolism and hypertension and vasculitis. However, upper airway disease was not one of the list. Two days beyond intubation she was extubated and discharged soon thereafter.

After discharge, she continued to suffer from recurrent episodes of severe dyspnea, hypoxia and breathing difficulties with any exertion, she sought medical advice in different clinics including general medicine, respiratory clinics and ER. However, she was always managed conservatively as a case of post COVID-19 respiratory and lung complications. After 3 months of this suffering, she came to an ENT clinic where upper airway disease was suggested for the first time, she presented with severe exertional dyspnea and harsh audible breathing sounds similar to biphasic stridor. She looked tired and was on a wheelchair, her throat and neck examination and fiberoptic laryngoscopic examination were all unremarkable. Computed tomography (CT) scan for the neck was done twice, the first of which did not show any stenotic segment. However, with a high index of suspension, a thin cut CT scan for the neck was repeated again and it showed severe subglottic and upper tracheal stenosis as shown in **Figure 1**.

The patient was scheduled for microlaryngoscopy and bronchoscopy which showed two areas of stenosis. The first one was grade 2 stenosis in the subglottic area (Cotton-Myer classification). The second one was grade 3 stenosis and it was 5 cm below the level of true vocal cords. Balloon and coblation-assisted dilatation was done during the procedure. After that, the patient had recurrence of the stenosis and she was prone to multiple surgeries to release the stenosis using

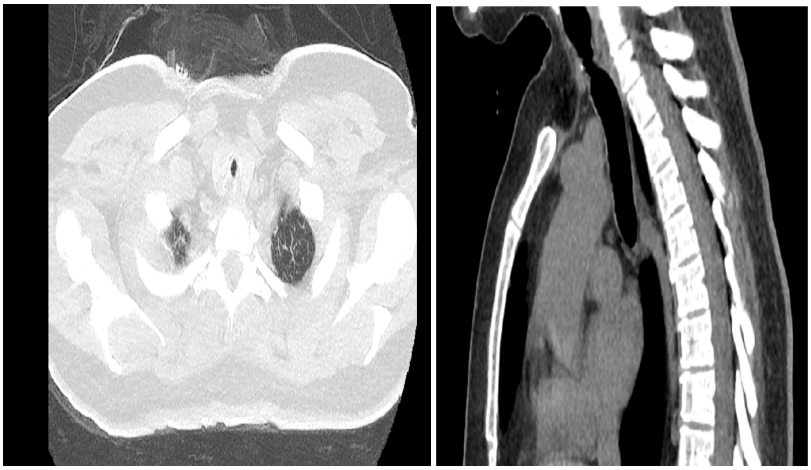

Figure 1. Computed tomography images of the chest and upper neck, showing focal area of irregular tracheal stenosis at the level of thyroid gland measuring around 1.1 cm in length, with minimum diameter of around 1 cm. (Right): Sagittal view, (left): Axial view.

balloon dilatation, coblation and laser. At this moment of writing this paper, the patient is planned for resection of the stenotic segment with end-to-end anastomosis.

Case 2

A 51-year-old male, who is a known case of type 2 diabetes and asthma, non-vaccinated for COVID-19, presented to ER with a 6-day history of persistent fever with vomiting and nausea associated with mild abdominal pain and cough. The patient had a positive history of contact with confirmed COVID-19 patient.

His vital signs were unremarkable except for fever (38.7 ˚C). Chest auscultation showed bilateral equal air entry with no added sounds. Lab tests showed CRP level (118) with lymphopenia. Chest x-ray showed bilateral peripheral ground glass appearance more on the right side. Abdominal ultrasound and CT scan did not show any pathology except basal peripheral ground glass opacification in the visible part of the lungs. COVID-19 was suspected and nasopharyngeal swab came positive, then the patient was admitted for further supportive treatment.

The patient's chest condition was deteriorating as he started to have a drop in O_2 sat and hypoxia. Finally, he developed respiratory failure which mandated ICU transfer then intubation and mechanical ventilation.

In the ICU, he remained intubated for 9 days before he had been extubated. After four days, he started to have severe respiratory distress for which he had been intubated again for two days. After the second extubation, his condition stabilized and he was transferred to regular ward and given the post COVID-19 supportive treatment. One week later, he was discharged home on O_2 nasal cannula.

In the following 2 weeks, he visited the ER and outpatient clinics 5 times with complaints of shortness of breath and easy fatigability with minimal movement in addition to hoarseness of voice, he was managed with nebulizers, cough syrup

and home inhalers. Then the patient visited the ER again with severe shortness of breath and drop in O_2 sat. Further investigations showed distal right sided pulmonary embolism for which he was admitted for 10 days for further medical treatment. During admission, the ENT team was consulted for his hoarseness of voice, a fiberoptic laryngoscopy revealed a picture of fungal laryngitis and was given antifungal treatment.

The patient's visits to ENT and respiratory doctors continued over the next month with complaints of shortness of breath and hoarseness of voice. Fiberoptic laryngoscopy was repeated by 3 different ENT doctors, and all showed bilateral mobile vocal cords with mild laryngeal edema and he was accordingly given antireflux medications.

LTS had not been suspected until the patient lastly came to the ER with severe distress and stridor. He looked tired, was on nasal O_2 cannula and sitting in a wheelchair, his head and neck and throat examination was unremarkable except the biphasic stridor which was increasing with any movement. CT scan for the neck was ordered and it showed LTS as appears in **Figure 2**.

The patient underwent microlaryngoscopy and bronchoscopy which showed grade 3 subglottic stenosis (Cotton-Myer classification), dilatation with coblation was performed. One month after the surgery, his symptoms recurred, so bronchoscopy was repeated, but this time the stenosis became more severe (grade 4) and was not candidate for dilation through the endoscope. Therefore, a tracheostomy was made, and postoperatively, the surgical option of resection of crico-tracheal stenotic segment with end-end anastomosis was offered to the patient but he refused it and preferred to stay on tracheostomy tube until this moment of writing this paper.

Figure 2. A Computed tomography images of the neck and chest, showing 1.5 cm long subglottic and upper tracheal stenotic segment that measures about 0.8 × 0.75 cm at its maximum anteroposterior and transverse dimensions respectively. (Right): Sagittal view, (left): Axial view.

3. Discussion

Post-intubation tracheal stenosis is a known complication of endotracheal intubation or tracheostomy. Factors that contribute to tracheal stenosis are cuff pressure, ETT size relative to the tracheal lumen, duration of intubation, hemodynamics during intubation, movement of the tube, age, sex, ETT material, and the use of steroids [7]. Prevention is possible to a high degree by use of large volume, low pressure cuffs [8].

The most important tools in the evaluation of laryngotracheal stenosis are microlaryngoscopy and flexible bronchoscopy in the operating room [9]. CT scan provides dimensions of the stenotic area for surgical planning purposes that can augment the laryngoscopy findings. Therefore, if there is concern for tracheal stenosis, then a CT with 0.5 mm cuts and three-dimensional (3-D) reconstruction is recommended [9]. In other words, the definitive diagnosis of LTS stenosis is done with bronchoscopy. A CT scan can give precise information regarding the site, level, and severity of obstruction. However, CT scan may sometimes miss the stenotic segment as occurred in the first case of this paper.

Management of stenosis located in the subglottic region is a major therapeutic challenge and is still controversial and there is no consensus about the best treatment strategy. The various forms of treatment described for LTS include laser, repeated endoscopic dilations, cryosurgery, prolonged stenting, laryngotracheal reconstruction and segmental resection with end-to-end anastomosis. However, cricotracheal/tracheal resection remains the most successful treatment for LTS to date [10].

Most common complications after any treatment modality are relapse and restenosis with recurrence within 1 - 3 months. Recurrence of stenosis differs according to treatment modality. The final success rate for airway stenosis varies between 40% and 80% based on the general condition of the patient, location and length of stenosis, and the preference of the surgeon for surgical technique [11].

In literature, many studies talk about post COVID-19 syndrome and its multi-organ effects including pulmonary, cardiac, renal, musculoskeletal, endocrine and neuropsychiatry [1] [2]. However, larygneal and tracheal effects are very rarely mentioned in this issue. Therefore, in this paper we highlight on LTS as an serious emerging sequelae of COVID-19 with both morbidity for patients and a further demand on healthcare resources. With the significant increase in tracheostomies and intubations performed during the COVID-19 pandemic, it is reasonable that a proportion of these will present with LTS in the future.

4. Conclusion

Lyrngotracheal stenosis is a serious complication after intubation and tracheostomy. With the significant increase in tracheostomies and intubations performed during the COVID-19 pandemic, it is reasonable that the number of patients who will end up with LTS increases. CDC is not including LTS as part of

post-COVID-19 syndrome. In this paper, we highlight the need to include LTS in this entity and publish that among health care workers owing to reduce the morbidity for those patients and the demand on healthcare resources, to avoid like what occurred to our two cases who were prone to much morbidity before LTS was diagnosed.

5. Key Clinical Massage

Laryngotracheal stenosis, due to prolonged intubation for COVID-19 pneumonia, is a considerable cause for severe respiratory distress after recovery from the acute phase of COVID-19 infection. Two years after the onset of this pandemic, more cases will evolve and they will need special care and management.

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Patient Consent

Consent was taken from patients to publish their cases.

Authors' Contributions

DA: collected the data,performed the literature search and prepared the manuscript.

MA: prepared and revised the manuscript.

RAH: prepared and revised the manuscript.

HA: revised the manuscript and submitted the paper.

All authors read and approved the final manuscript.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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