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CASE REPORT

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Ludwig's angina in a COVID positive patient—An atypical case report

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Abstract

The management of odontogenic infections is established and several guidelines have been proposed for its effective treatment. Outbreak of COVID-19 has posed serious challenge in the management of odontogenic infections, further complicated by immunocompromised status of the patient, where adequate evaluation and prompt attention is mandatory to avoid untoward consequences. Ludwig's angina being a life threatening infection by itself, association or simultaneous presentation with COVID-19 infection can add further complexity in the management for a maxillofacial surgeon. Here we present a case report of Ludwig's angina in a patient who tested positive for Coronavirus infection and how we treated her successfully.

KEYWORDS

COVID-19, extraction, Ludwig's angina, odontogenic infection

1 | INTRODUCTION

Ludwig's angina, a life threatening infection that is defined as a severe cellulitis with involvement of bilateral submental, submandibular and sublingual spaces, can spread into parapharyngeal spaces and cause laryngeal edema leading to airway obstruction.¹ Odontogenic infection arising from lower molars particularly, is the most common etiology in the occurrence of Ludwig's angina whereas the nonodontogenic causes include sialadenitis of submandibular gland, mandibular fracture and traumatic injury to the floor of mouth.^{2,3} Early surgical drainage supported by medical management being the mainstay of treating Ludwig's angina, decreases the stress on airway and provides better antibiotic penetration.⁴ Antibiotic sensitivity testing and culture can aid in administration of culture specific antibiotics. Ludwig's angina requires a complex multidisciplinary approach and this may be further complicated if the patient is positive for COVID-19 because of the need for isolation and extensive care to avoid sepsis and airway collapse. Pneumonia caused by COVID-19 can further complicate the existing airway obstruction due to Ludwig's angina.⁵

Management of Ludwig's angina with simultaneous COVID-19 infection requires a complex protocol for complete recovery which must involve all the treatment modalities to manage the infection, airway, existing comorbidities and COVID-19 associated symptoms.

2 | CASE REPORT

A 26-year old female reported to the emergency department with swelling in bilateral sublingual and submandibular region since three days. The patient had pericoronitis with respect to lower right third molar (48), one week back for which she was prescribed antibiotics and analgesics from a nearby dental clinic. However, the

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FIGURE 1 Radiograph showing foci of infection 16, 36, 46 root stumps and carious 26, 37

infection did not subside and the swelling which was initially pea sized gradually progressed to the present size 7×8 cms (Anteroposterior x Transverse dimension). The swelling was not fluctuant and extended from one side of the body-angle region of the mandible to the other side and from the submental region anteriorly to the hyoid bone inferiorly. Intraorally, the consistency was brawny with raised floor of mouth and tongue displaced towards the palate slightly. Orthopantamograph (OPG) showed root stumps 16, 36 and 46, carious 26, and 37 (Figure 1) and computed tomography (CT) of neck showed no parapharyngeal involvement. She was straight away diagnosed to have Ludwig's angina based on the clinical presentation. At the emergency department, blunt bilateral submandibular incisions were placed under local anesthesia to decompress the collection and sample was sent for culture and sensitivity testing. Empirical antibiotic therapy was initiated with Cefotaxime 1 g intravenously thrice a day. As the patient developed signs of upper respiratory infection, she was shifted to Infection ward and kept under observation. Subsequent Contrast Enhanced CT (CECT) neck and thorax after 24 h showed extension into the pharyngeal space (Figure 2) with consolidations in both the lungs and features pointing towards a possible COVID-19 infection. The patient tested positive for COVID-19 by Antibody testing and was then isolated in the designated COVID-19 ward. As there was no significant improvement, the bilateral submandibular incisions were joined with a single long rubber drain following all the COVID-19 protocols. Elevated D-Dimer level $(0.75\mu g/ml)$ prompted us to start Enoxaparin 40 mg subcutaneously once daily, to prevent intravascular thrombosis. C-Reactive Protein (CRP) and Procalcitonin levels were increased to 107 mg/L and 6.14 ng/ml respectively. On the third day, patient had oxygen saturation fall (90%) that did not improve even with oxygen support. Oral Favipiravir (1800 mg twice a day on first day, followed by 800 mg twice a day for seven days), Etofylline 84.7 mg + Theophylline 25.3 mg 2 ml intravenous administration, and Salbutamol inhalation were started on the third day. Swab culture result showed growth of Alpha hemolytic streptococcal colonies susceptible to Gentamicin,



FIGURE 2 The CECT axial view showing involvement of pharyngeal space (marked by blue arrow) and submandibular spaces (marked by yellow arrow)

persuading us to change the antibiotic to a culture specific one. By sixth day, the right buccal space and masticatory space were also involved (Figure 3) with trismus clinically and her general status further declined. Hence, patient was shifted to intensive care unit (ICU) and planned for tracheostomy if the fall in oxygen saturation did not improve. Extraction of 16, 26, 36, 37, 46 and 48 were done and collection was drained from the right buccal and pterygomandibular space under local anesthesia. Re-exploration of the bilateral submandibular region was done followed by local irrigation of 100 ml metronidazole solution and active mouth opening with Ferguson mouth gag.

Though the patient was shifted to ICU, tracheostomy was not required as the oxygen saturation improved (96%) after extraction of involved teeth, mouth opening and re-exploration were performed promptly. Frequent monitoring of vital signs, copious local irrigation of antibiotic solution (200 ml metronidazole solution twice a day, using 20 ml syringe without needle), antiseptic (amorphous hydrogel with colloidal silver) dressing, follow up with CRP and Arterial Blood Gas analysis were done.



FIGURE 3 Status of patient on day six with Ludwig's angina showing spread of infection to right buccal space clinically

Active mouth opening once a day daily, was performed with mouth gag during our daily visits and the patient was encouraged for ice cream stick exercise in between, on her own. Patient showed good response by the eighth day.

The patient was discharged after testing negative for coronavirus and drain removed on the fifteenth day. At the time of discharge, the CRP and procalcitonin levels were 10 mg/L and 1.11 ng/ml respectively. Patient's general condition improved and started taking solid diet.

3 | DISCUSSION

Ludwig's angina being a rarely encountered but wellknown emergency, begins as a localized odontogenic infection commonly and spreads rapidly to involve the submaxillary and pharyngeal spaces. Airway compromise and posterior displacement of tongue are of concern in this condition warranting immediate attention.⁶ The COVID-19 infection caused by Severe Acute Respiratory Syndrome Coronavirus-2 (SARS-CoV-2), primarily affects the lung, producing breathing difficulties and may lead to various complications.⁷ The unforeseen damages produced by the COVID-19 pandemic have affected the presentation of odontogenic infections due to inaccessibility and lack of provisions for proper patient care during the lockdown period.⁸ Simultaneous COVID-19 infection can complicate the management of Ludwig's angina by worsening pneumonia, pulmonary embolism or septic shock.^{10,11} Blind oral intubation is not recommended in Ludwig's angina due to fear of rupture of the abscess. Elective tracheostomy is a useful surgical intervention in preventing the impending airway emergency although it may be difficult in certain cases due to the distortion of normal anatomy of neck.¹² Therefore, prompt management of airway and surgical decompression are crucial in the management of Ludwig's angina.

SARS-CoV-2 accessing the endothelial tissues via spike protein envelope, damages the endothelium and alters the coagulation process, thus promoting thrombosis.¹³ Cytokine storm in COVID-19 with increased levels of Interleukins—1, 6, 8 and tumor necrosis factor (TNF)— α are shown to be associated with thrombo-embolic events.¹⁴ The exact clarification for thrombotic events to occur in COVID-19 has not been provided.¹³ However, studies have shown the need for prophylactic anticoagulants monitored by D-dimer levels to avoid intravascular coagulation episodes.¹⁵

Despite clinical presentation being the primary clue for diagnosis of Ludwig's angina, radiological evaluation with CT or magnetic resonance imaging (MRI) provides an accurate picture of the underlying spread of disease and involvement of vascular structures. In the pre antibiotic era, the mortality of Ludwig's angina was high with airway distress being the underlying cause of death of the affected patients.¹⁶ With newer antibiotics, diagnostic methods and surgical techniques, prognosis of Ludwig's angina has improved greatly.¹ Overall, the approach must be step-wise with careful monitoring and evaluation to avoid airway emergency.

Surgical decompression supported by culture sensitive antibiotics still remains the gold standard protocol. Drainage must be done with proper personnel protective measures and in isolation.9 Unless extraction of the focus tooth is done, the septic focus remains unaddressed.¹⁸ Guidelines mandate extreme caution while performing transoral and transnasal procedures after ascertaining the COVID-19 status of the patient and advice use of personal protective equipment (PPE) and powered airway purifying respirators if the patient is found positive for the virus.⁹ The use of routine handpiece method for surgical extraction is not recommended due to risk of aerosol spread of COVID-19 and hence we opted for age-old chisel and mallet technique for trans-alveolar extraction of 48.¹⁹ Surgical decompression must be carried out by incision and drainage of the involved spaces. If unattended or delayed, involvement of pharyngeal and mediastinal spaces can occur insisting aggressive management.

Routine laboratory investigations may not sufficiently diagnose sepsis syndrome and hence procalcitonin levels are recommended to be included in the follow up check list.¹⁷ Meticulous follow up is absolutely necessary for a successful treatment. As Ludwig's angina complicated by COVID-19 infection demands increased attention and

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immediate decisions, this brief presentation being the first to report one such scenario to our knowledge, calls for anticipation of such encounters with swift response to successfully manage them.

4 | CONCLUSION

A multidisciplinary and step-wise approach must be adopted to treat the simultaneous infections. Early identification and treatment of odontogenic infections during this COVID-19 pandemic is important to prevent deterioration. No specific treatment outline is available in literature at present to manage Ludwig's angina in a patient with COVID-19 infection. Unless paramount care is given to avoid sepsis and airway emergency, the management of these cases can be a dreadful experience. Hence, this case report is aimed at emphasizing the need for utmost care in Ludwig's angina with simultaneous COVID-19 infection, anticipating airway distress, sepsis or any complications of COVID-19 and be prepared to address them.

DECLARATION

FUNDING

The authors have not received any external source of funds/grants.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

ETHICS STATEMENT

The Institutional Review Board of our hospital did not demand Institutional Ethical Clearance for this case report. The patient has signed an informed written consent form for publishing this case report and is in compliance with the Declaration of Helsinki. Complying to the strict Hospital guidelines, clinical and intra oral pictures of patient procedures and follow up were not taken inside the COVID-19 ward and ICU.

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