Successful treatment of intubation-induced severe neurogenic post-extubation dysphagia using Pharyngeal Electrical Stimulation in a COVID-19 survivor: a case report

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Case Report

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Abstract

**Background** A significant portion of critically ill patients with COVID-19 are at high risk of developing ICU-acquired swallowing dysfunction (neurogenic dysphagia) as a consequence of requiring prolonged mechanical ventilation. Pharyngeal Electrical Stimulation (PES) is a simple and safe treatment for neurogenic dysphagia. Previously it has been shown that PES can restore safe swallowing in orally intubated or tracheotomised ICU-patients with neurogenic dysphagia following severe stroke. We report the case of a patient with severe neurogenic post-extubation dysphagia (PED) due to prolonged intubation and severe general muscle weakness related to COVID-19, which was successfully treated using PES.

**Case Presentation** A 71-year-old female patient with confirmed SARS-CoV-2 infection developed neurogenic dysphagia following prolonged intubation in the ICU. To avoid aerosol generating procedures, her swallowing function was evaluated non-instrumentally as recommended by recently published international guidelines in response to the COVID-19 pandemic. Her swallowing function was markedly impaired and PES therapy was recommended. PES led to a rapid improvement of the post-extubation dysphagia (PED), as evaluated by bedside swallowing assessments using the Gugging Swallowing Screen (GUSS), Dysphagia Severity Rating Scale (DSRS) and diet screening using the Functional Oral Intake Scale (FOIS). The improved swallowing, as reflected by these measures, allowed this patient to transfer from the ICU to a non-intensive medical department five days after completing PES treatment.

**Conclusions** PES treatment contributed to the restoration of a safe swallowing function in this critically ill patient with COVID-19 and ICU-acquired swallowing dysfunction. Early clinical bedside swallowing assessment and dysphagia intervention in COVID-19 patients is crucial to optimise their full recovery. Pharyngeal Electrical Stimulation may contribute to a safe and earlier ICU discharge of patients with ICU-acquired swallowing dysfunction. Earlier ICU discharge and reduced rates of re-intubation following PES can help alleviate some of the pressure on ICU bed capacity, which is critical in times of a health emergency such as the ongoing COVID-19 pandemic.

**Background**

Following the world-wide spread of the novel coronavirus infection (SARS-CoV-2), which first emerged in China in December 2019, the WHO declared the coronavirus disease 2019 (COVID-19) outbreak a pandemic on the 11th of March 2020. The percentage (9-32%) of severely or critically ill COVID-19 patients admitted to intensive care units (ICU) were diverging in the different affected countries (1-4). Overall, 20 to 40% of patients required mechanical ventilation by the time of their ICU admission in the USA (5).

Prolonged ICU stay - especially prolonged duration of mechanical ventilation coupled with sedation and bed rest - is associated with neurological disorders due to a reduction in afferent sensitivity; consequently, there is a high risk of dysfunction of the swallowing musculature, which can persist for months or years.
after ICU discharge (6). COVID-19 can also manifest with neurologic symptoms (7), myopathia as well as myositis (8), which aggravate PED. Indeed, the trauma caused by the endotracheal tube is considered one of the main causes of ICU-acquired dysphagia (9). Post-extubation dysphagia (PED) is defined as swallowing difficulty after extubation. In a systematic review including nine clinical studies analysing 775 ICU-patients after oral endotracheal intubation, 49% of patients presented with PED (9). A large prospective observational trial (DYnAMICS) evaluating dysphagia in 1304 mechanically ventilated, unselected ICU-patients, reported a PED incidence of 18.3%. Moreover, in that study, among the 90 patients discharged from hospital, 58 (64.4%) showed persistent swallowing dysfunction (10). Dysphagia complications following prolonged hospitalization are well-known and associated with a high risk of malnutrition and dehydration, decreased health-related quality of life, as well as an elevated occurrence of aspiration pneumonia. Aspiration pneumonia is a frequent cause of hospital re-admission and mortality in ICU-patients (11, 12). The consequently repeated need of antimicrobial therapy is a breeding ground for antimicrobial resistance. Despite its frequent occurrence in intubated patients and significant morbidity, there is a lack of awareness of PED and its consequences.

Currently, three different therapeutic options exist for treatment of oropharyngeal dysphagia: i) patient-specific variation in dietary texture, ii) postural changes/compensatory manoeuvres, and iii) medical devices delivering personalized neuromuscular stimulation to improve swallowing function by treating the afferent sensory pathways involved in the pharyngeal impairment (13). The Phagenyx system (a medical device CE-marked since 2012 from Phagenesis Ltd., Manchester, UK) has been designed to treat the symptoms of swallowing disorders by stimulating sensory nerves in the oropharynx through directed electrical pulses. The Pharyngeal Electrical Stimulation (PES) system uses a nasogastric feeding tube-like stimulation catheter incorporating two specially designed electrodes (Figure 1). Through communication with the patient, the healthcare worker finds an individually adjusted stimulation intensity for optimised PES. Typically, stimulation is delivered using a current intensity range between 1-50 mA, a stimulation frequency of 5 Hz, and a pulse width of 200 μs. Each period of PES usually lasts for ten minutes. Three treatment sessions may be given on several consecutive days to patients with neurogenic dysphagia (14, 15). PES therapy has been used to safely and successfully treat neurogenic dysphagia in patients presenting with multiple medical conditions: in orally intubated ICU-patients (15), in tracheotomised severely dysphagic stroke patients (14, 16, 17), in patients with brain injury (18) and patients suffering from multiple sclerosis (19).

We describe the case of a COVID-19 patient presenting with severe general muscle weakness and ICU-acquired swallowing dysfunction, who - despite her viral infection and prolonged ICU stay - was successfully treated with PES (post-extubation). This enhanced safe swallowing, allowed the patient to resume a more normal nutritional intake, avoided a re-intubation or tracheostomy and permitted therefore a stable ICU discharge.

**Case Presentation**

**Patient's relevant demographic details and medical history**
The 71-year-old female patient has a medical history significant for bilateral hydronephrosis, bilateral subpelvic ureteral obstruction, pneumococcal pneumonia (2008), and restless legs syndrome. She presented to the urology department of a partnering public hospital in Vienna (Austria) on April 3rd, 2020 with complaints of abdominal pain and a dry cough (see Figure 2 for a schematised summary of patient hospitalisation timeline). Upon initial assessment, the patient demonstrated partial respiratory insufficiency with a peripheral SpO2 of 86%. On Day 3 after admission, a chest x-ray was performed, which showed a typical bilateral viral pneumonia. On Day 4, the respiratory insufficiency deteriorated quickly (10 L/minute O2 required to maintain SpO2 >90%), so the patient was transferred to the intensive care unit (ICU) and intubated. Laboratory tests revealed a lymphopenia and C-reactive protein (CRP) elevation, both typical biomarkers of the COVID-19 disease (1,2). After confirmation of SARS-CoV-2 through PCR out of a nasopharyngeal swab, the patient was transferred (mechanically ventilated) to our ICU, being the main ICU in charge of managing COVID-19 patients in Vienna (IV Med. Dept., Kaiser Franz Josef Hospital). Prior to hospitalization, the patient was living independently and was safely managing a regular solid and thin liquid diet with no history of dysphagia.

**Antiviral and antibiotic therapies**

Hydroxychloroquine was initiated on Day 7 after admission as an antiviral treatment for COVID-19 and discontinued on Day 8 due to prolongation of QTc time. Subsequently, the patient received standard supportive care for a viral pneumonia. Following the initial diagnostic PCR on Day 4, PCR for SARS-CoV-2 was performed out of tracheal secretions and nasopharyngeal swabs twice a week (as long as secretions could be aspirated). On Day 21, the PCR out of the nasopharyngeal swab was only weakly positive, while tracheal secretion was still clearly positive. On Day 49, the nasopharyngeal swab sample was negative for the first time, at that time tracheal secrete could not be obtained for testing.

On initial admission, ampicillin/sulbactam was started empirically. After transfer to the COVID-19 ICU, the antibacterial treatment was switched to piperacillin/tazobactam, which was stopped after seven days of treatment in the absence of evidence of a bacterial infection. On Day 22, increased putrid pulmonary secretions were observed, and a PCR test was conducted on the patient's sputum (pneumonia panel plus, Biomerieux) targeting 33 different kinds of bacteria and viruses. *Klebsiella pneumoniae* was detected and antimicrobial therapy was started with cefepime. The patient's general condition worsened on Day 27; as she also developed elevated inflammation parameters (leukocytes and CRP), the antibiotic therapy was escalated to meropenem. The patient's condition thus improved, and the therapy was de-escalated to ceftazidime, which was further administered between Day 32 and Day 36.

**Respiratory support**

The patient was mechanically ventilated via endotracheal tube for twelve days from Day 4 until Day 16 of the hospital stay. Following extubation, the patient received non-invasive ventilation (NIV) with a full-face mask. The next day, she was transitioned to high flow nasal cannula (AIRVO2). However, on Day 25, the patient's general health condition deteriorated rapidly. Hence, continuous NIV with full-face mask was
necessary again until Day 29; after which high flow nasal cannula was restarted. On Day 35, the patient was finally put on oxygen insufflation through low flow nasal cannula and weaned to breathing room air in the following days.

**Baseline dysphagia assessments**

Speech and language therapy services were consulted following endotracheal extubation on Day 16 to assess the swallowing function. These assessments were performed initially at bedside; they revealed oral and suspected pharyngeal swallowing dysfunction manifest as poor saliva management with observed drooling, suspected reduced hyolaryngeal elevation as judged via palpation during volitional swallowing, and aphonia. Consequently, nil by mouth was recommended by the speech and language therapist (SLT) (Dysphagia Severity Rating Scale score (20), DSRS = 12; Functional Oral Intake Scale score (21), FOIS = 1). Due to the COVID-19 pandemic and restrictions on the conduct of aerosol generating procedures, the SLT was unable to complete an instrumental assessment of the swallow, such as a fibreoptic endoscopic evaluation of swallowing (FEES) or videofluoroscopy (VFS), to assess more directly the patient's oropharyngeal swallowing function. Hence, dysphagia evaluations were limited to clinical bedside screening and non-invasive assessment tools.

On Day 18, SLT performed a clinical bedside swallow evaluation using the Gugging Swallowing Screen (GUSS) (19); the patient reached a total score of 6 out of 20, indicating the presence of severe dysphagia with a high risk of aspiration. The diet recommendation of nil by mouth (DSRS = 12; FOIS = 1) was maintained. During the screening, the patient continued to demonstrate poor management of oropharyngeal secretions with minimal reflexive swallowing and required frequent suctioning. SLT administered a trial of thickened water, which was followed by an unproductive cough response, further impacted by continued generalized muscle weakness, a typical feature seen in COVID-19 patients. On Day 20, the swallow function was re-assessed via GUSS and the patient received a score of 7 out of 20, again indicating severe dysphagia with a high risk of aspiration with continued recommendation of nil by mouth (DSRS = 12; FOIS = 1); these represented the initial GUSS, DSRS and FOIS baseline values prior to PES start on Day 22 (Figure 3). Informally, the patient was observed having increased difficulty managing secretions, required more frequent suctioning and had an oxygen saturation of 86%.

Subsequently, on Day 20 and Day 26, the patient developed aspiration pneumonia requiring antibiotic treatment.

**PES Treatment**

Due to persistent severe dysphagia, two cycles of PES therapy were performed: a first cycle between Day 22 and Day 26 (5 sessions) and a second cycle on Day 33 and Day 34 (2 sessions). Each daily session lasted ten minutes. This patient was the first COVID-19 patient receiving PES in our department. The delayed start of PES therapy was due to unfounded ambiguities within the team concerning hygiene matters. Stimulation parameters (current, mA) are described in Table 1.

| Table 1: Pharyngeal Electrical Stimulation (PES) current intensities |
Following the first three sessions of PES treatment, swallowing improvements were observed, including improved secretion management, and increased reflexive swallowing; however, the patient’s medical condition deteriorated on Day 25 with increasing inflammation parameters. The persistent extreme muscle weakness caused a moribund state. Hence, on Day 27, the behavioural therapy, including PES, was paused. Because of stress related to the necessary NIV-treatment, the patient was sedated until Day 29 when clinical improvement was observed (normal heart and breathing rates, good vigilance). On Day 32, the patient’s medical status and vigilance were stable enough to restart standard behavioural treatment. Therefore, a second PES cycle was performed on Day 33 and Day 34 with patient receiving seven stimulation sessions in total across two treatment cycles (Table 1).

| Date of PES | PES cycle 1 | PES cycle 2 |
|-------------|-------------|-------------|
|             | Day 22      | Day 23      | Day 24 | Day 25 | Day 26 | Day 33 | Day 34 |
| Stimulation (mA) | 48         | 15         | 22     | 19     | 13     | 16     | 12     |

Treatment Outcome

On Day 38, four days following the final PES treatment session, SLT re-evaluated swallowing and noted a GUSS total score of 15 out of 20, describing light dysphagia with a low risk of aspiration—a significant improvement from baseline GUSS scoring of 7 out of 20 (Figure 3). Due to these functional improvements, the patient’s diet was advanced from nil by mouth (DSRS = 12; FOIS = 1) to mushy-homogeneous solid food (International Dysphagia Diet Standardisation Initiative (22), IDDSI = 4) and thickened fluids (IDDSI = 3) administered in teaspoon amounts using a compensatory chin tuck posture for increased swallow safety (DSRS = 9; FOIS = 2) (Figures 4 and 5). On Day 41, GUSS was re-assessed with additional improvement noted as patient scored 17 out of 20 indicating light dysphagia with a low risk of aspiration; the patient’s diet was advanced to semisolids with soft consistency foods (e.g. white bread, IDDSI = 5) and concentrated fluid (IDDSI = 2), given while the patients was in an upright position in a transverse bed during meals (DSRS = 6; FOIS = 4) (Figures 4 and 5). It is important to note that due to the patient’s overall improved swallowing function and safety, she was discharged to a non-intensive medical department on Day 39, and the feeding tube was removed on Day 41. Final post-treatment GUSS was completed on Day 56 and revealed a total score of 19/20. The patient’s diet was upgraded to soft solid foods (IDDSI = 6) and thin liquids (IDDSI = 0) (DSRS = 3; FOIS = 6) (Figures 4 and 5).

On May 27th, 2020 (Day 54) the patient was transferred to acute geriatrics for further intensive physiotherapeutic mobilisation with the objective of returning to living independently at home again.

Discussion And Conclusions

We are presenting the first case report of a COVID-19 patient with ICU-acquired PED showing full recovery of swallowing function following PES therapy, and importantly, in absence of any PES-related adverse events. COVID-19-associated symptoms need to be taken in consideration in the treatment of dysphagic
patients, as these patients seem to be at increased risk for healthcare-associated infections. Indeed, bacterial co-infections, as observed in this patient, seem to be a typical characteristic of severe and critical forms of SARS-CoV-2 infection (23), as well as severe muscle weakness (24).

PED, which has been reported by up to 62% of patients with severe COVID-19 requiring mechanical ventilation, is one of the mid- to long-term sequelae (11). Critically ill COVID-19 dysphagic patients seem to be particularly at higher risk of aspiration and subsequent aspiration pneumonia; indeed, brain areas, peripherical nerves and muscles, which are responsible for normal deglutition, are often impaired as a consequence of the COVID-19 disease (12, 25). The swallowing dysfunction, observed in the previously dysphagia-nave patient reported here, is most likely the result of endotracheal tube trauma; a commonly observed problem in cases of intubation in an emergency setting (10). Our observations, as reported in a recent publication (26), also suggest that there is a direct neurological component involved in the pathology of COVID-19 patients. Further research will nevertheless be needed to understand the full extent of this novel coronavirus disease and its consequences.

Of note, a high stimulation level was required on the first day of PES, showing the lack of neuromuscular sensitivity by this patient following her prolonged (twelve days) intubation; such reduced local sensitivity has been previously described in ICU-acquired dysphagic patients (12). From the second stimulation day, a more normal range of stimulation values were used according to patient responses, showing a recovery of sensibility after a single ten minutes PES treatment session. PES can be delivered whilst patients are still intubated as well; in this case, stimulation threshold can be deduced from more autonomic cues from the lightly sedated patients (RASS-1) such as sweating, mimics, increased blood pressure or stress. A recent pilot study by Koestenberger et al. showed a significantly earlier improvement in swallowing after PES treatment in orally intubated ICU-patients compared to those stimulated following extubation; suggesting a faster recovery of dysphagia when PES is performed sooner (15). Moreover, patients receiving PES-treatment had lower prevalence of pneumonia and frequency of re-intubation than patients without PES stimulation. Had we started PES therapy during intubation in this patient, we may have accelerated her recovery by minimising the consequences of dysphagia.

During our patient’s ICU stay, the ongoing therapeutic measures had to be postponed – including PES dysphagia therapy for six days - due to the de novo sedation of the patient because of the deterioration of her general medical state; however, thanks to the first cycle of PES, a re-intubation or tracheostomy of the patient has been thus avoided.

At the beginning of the COVID-19 outbreak, the priority of ICUs was simply to keep patients alive, and therefore, no other therapeutic procedures except those associated with intensive care were performed. As time progressed, medical staff gained experience and came slowly back to normality. A balance had to be found between the limited exposure of medical staff and the rights of critically ill COVID-19 ICU-patients for an optimal health-related quality of life and the full program of medical care they needed. In this sense, a recently published WHO guideline recommends continuous rehabilitation interventions for patients with severe COVID-19 (27).
For ICU-patients, systematic bedside screening for PED - for which a feasible and pragmatic approach has been already published (12) – as well as rehabilitation measures of neurogenic dysphagia should be implemented into clinical protocols. To minimize risks of viral exposure through aerosol emissions during dysphagia therapy, medical staff should wear adequate PPE for procedures on suspected or confirmed COVID-19 cases (28, 29). This will allow early dysphagia assessment and the subsequent implementation of personalized treatment (12). COVID-19 survivors in the acute, sub-acute and long-term phases of care have specific rehabilitation needs; PES therapy can be used to facilitate early recovery, further transfer of dysphagic patients to non-intensive case rehabilitation facilities and contribute overall to shorten hospital length of stay.

Clinical management guidelines related to dysphagia therapy, as well as international intensive care societies like for instance the GARI in Austria, recommended in their recently published position statements to restrict aerosol generating procedures on COVID-19 positive or suspected patients in order to avoid contamination risk for healthcare personal (26, 30). Consequently, only non-instrumental bedside examinations were performed to assess dysphagia in potentially infected ICU-patients (26-28). Although instrumental assessments in COVID-19 positive cases are currently limited, an optimal rehabilitation of PED following prolonged ICU stay is feasible; the use of SLT interventions or equipment-based therapies like PES leads to an effective and early management of dysphagia (31). A holistic approach within an intensive care multidisciplinary team is needed to optimally take care of acute and early post-acute neurogenic dysphagic patients. In absence of any established standard treatments for dysphagic COVID-19 patients, PES may facilitate early discharge of COVID-19 patients to non-intensive care units and reduce the risk of ICU re-admission. As we have unfortunately learned from the ongoing COVID-19 pandemic, maintaining adequate ICU efficiency and bed capacity is a critical factor in preventing the healthcare system from being overwhelmed.

**Abbreviations**

AIRVO: humidified high flow system

DSRS: Dysphagia Severity Rating Scale

FEES: fibreoptic endoscopic evaluation of swallowing

FOIS: Functional Oral Intake Scale

GUSS: Gugging Swallowing Screen

IDDSI: International Dysphagia Diet Standardisation Initiative

NIV: non-invasive ventilation

PED: post-extubation dysphagia
PES: Pharyngeal Electrical Stimulation
RASS: Richmond Agitation-Sedation Scale
VFS: videofluoroscopy

Declarations

Ethics approval and consent to participate

Ethics approval was not applicable to this case report. The patient gave written consent for data collection.

Consent for publication

The patient gave written consent for publication.

Availability of data and materials

The datasets used and analysed are available from the corresponding author on reasonable request.

Competing interests

None of the authors have competing interests to declare.

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Authors' contributions

All authors contributed equally to this work; all of them read and approved the final manuscript.

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Authors' information

All co-authors are actively working as clinicians at the ICU department and were actively involved in the case. All authors read and approved the final manuscript.

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