

## Tapia syndrome: an Unusual Complication to Recognize at the Time of the COVID-19 Pandemic

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### Abstract

Guidelines for acute respiratory distress syndrome (ARDS) management include sedation, neuromuscular blockade for protective ventilation and the need for the prone position. This technique can lead to neurologic sequelae like the Tapia syndrome following the patient position. We reported two ARDS COVID-19 patients with Tapia syndrome due to prone position. Both patients, a white European 57 year-old female and a Black African 50 year-old male, were intensive care unit (ICU) admitted for ARDS due to COVID-19. They were orotracheal intubated and mechanical ventilated for respectively 23 and 68 days with 7 and 4 sessions of prone position. Both were tracheotomized during their ICU stay. We observed during the respiratory weaning, swallowing difficulties associated with a tongue deviation during protrusion and paralysis of the ipsilateral vocal cord suggesting a Tapia syndrome. Prone position for the treatment of hypoxic ARDS COVID-19 patients with lateral flexion of the head could induce a compression on the lower cranial nerves and their collateral branches and lead to the Tapia syndrome with long neurologic sequelae: swallowing disorder, dysarthria and dysphagia due to difficulty to move the tongue with paralysis of the vocal cord. Because mechanical ventilation on prone position is a cornerstone of the treatment of hypoxic COVID-19 patients, the number of patients with Tapia syndrome may increase in the future.

**Keywords:** ARDS COVID-19; Prone Position; Tapia Syndrome; Safety; Intensive Care

**Abbreviations:** ARDS- Acute Respiratory Distress Syndrome; ICU- Intensive Care Unit

### Introduction

Severe hypoxemia with ARDS requiring mechanical ventilation is the principal cause of ICU admission in patients with SARS-CoV-2. Guidelines for protective mechanical ventilation include small tidal volume with a driving pressure below 15 cmH<sub>2</sub>O, use of sedation, neuromuscular blockade and prone position also in awake patients [1-3]. A series of epidemiological studies has confirmed that oxygenation improves with prone position in 60-80 % of COVID-19 patients [4]. Nevertheless, this technique has some side effects. Here, we described two cases of patients managed by prone position for hypoxemia due to ARDS COVID-19. They developed Tapia syndrome secondary to a stretching of the nerves during lateral head flexion.

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## Case Description

### Case 1

A white European 57 year-old female treated for arterial hypertension, was admitted for hypoxemia due to COVID-19 infection. She was treated by non-invasive ventilation but finally was intubated after 24 hours. Severe ARDS (PaO<sub>2</sub>/FiO<sub>2</sub>: 57) required 4 sessions of prone position during ICU stay. Tracheostomy has been delayed after 25 days due to several episodes of ventilatory acquired pneumonia, venous thromboembolism and pneumothorax due to barotrauma. After 50 days, she leaves ICU to the department of rehabilitation with oxygen on tracheotomy. During ICU stay, we have observed an ICU acquired neuromyopathy with palsy of the left arm and abnormal swallowing. Fifteen days after ICU discharge, electromyographic exploration confirmed severe lesions of the left brachial plexus with absence of spontaneous activity on the cubital and median nerves. This could be explained by the stretching of the plexus during the prone position as reported [5, 6]. Investigations of the swallowing showed a paralysis of the right vocal cord revealing right recurrent laryngeal nerve paralysis and a right deviation of the tongue during protrusion. No evidence of other cranial nerves was observed. These neurologic alterations are known as Tapia syndrome with concomitant lesions on the X and XII nerves due to prolonged lateral head flexion like in prone position. After three months of rehabilitation and regular logopedic exercises, the patient status improves with better swallowing and the gastrostomy feeding tube could be removed. Unfortunately, the protrusion of the tongue as well as the paralysis of the vocal cord are still present making dysphonia.

### Case 2

A Black African 50 year-old male, with a medical history of type II diabetes, was admitted for dyspnea due to COVID-19 infection. He quickly required orotracheal intubation and mechanical ventilated during 21 days with 7 sessions of prone position ventilation. He was extubated but reintubated the same day for hypoxemia probably linked to swallowing difficulties. A tracheotomy was performed after 23 days of mechanical ventilation. During the ventilatory weaning, we noted swallowing difficulties associated with a tongue deviation toward the left side during protrusion due to a left hypoglossal nerve paralysis (Figure 1A).

There were no other deficient nerves. A nasofibroscopy showed paralysis of the left vocal cord due to a left recurrent laryngeal nerve paralysis suggesting a Tapia syndrome. After two months of intensive logopedic treatments, these deficits slightly improve (Figure 1B).



**Figure 1A:** At ICU discharge, the patient shows a tongue deviation during protrusion associated with a paralysis of the vocal cord.



**Figure 1B:** After two months, neurological deficits slightly improve.

## Discussion

Since first cases described in end of 2019, mortality of ARDS COVID-19 patients remains high, especially in patients on mechanical ventilation and despite several procedures to limit ventilator-induced lung injury. Initial management of a patient with ARDS included lung protective ventilation strategies prioritize lower tidal volumes, higher positive end-expiratory pressure, and limiting driving pressure [1, 2]. To obtain this optimal ventilatory management and limiting oxygen consumption, sedation and neuromuscular blockade were also used [1, 2]. Since the Prone Severe ARDS Patients (PROSEVA) study [7], which showed a significant effect with prone position on mortality at 28 and 90 days in patients with severe ARDS (PaO<sub>2</sub>/FIO<sub>2</sub> < 150 mmHg), prone position has been recommended as an adjunctive therapy in these patients but has only been very few used. In the LUNG SAFE study, which reported the epidemiology and care in

ARDS patients, only 187/2377 (8 %) for all ARDS patients and 119/729 (16 %) in severe ARDS patients were on prone positioning as adjunctive therapy [8]. The COVID-19 pandemic has increased the use of prone position and even in awake patients on non-invasive ventilation [3, 9]. Given the number of patients in hospitals, some centers have even developed teams dedicated to this practice [10]. Nevertheless, this technique is not any risks and complications were described including neurologic complications. In a recent review, Gonzalez-Seguel et al. reported 8.1 % of peripheral nerves (brachial plexus, ulnar, radial, sciatic and median nerves) [6]. We also report a case of brachial palsy but in these two cases another nerve injury due to prone position. Tapia's syndrome is an entity characterized by unilateral paralysis of the tongue and vocal cord due to lesion of both recurrent laryngeal and hypoglossal nerves [11- 17]. Tapia syndrome is due in compression or stretching of these nerves on their extracranial course. Lateral head flexion during prone position have probably caused this injury. Symptoms of Tapia syndrome includes dysphonia, tongue deviation towards the affected side, impaired mobility of the tongue and difficulty in swallowing. Actually, few other cases of patients with Tapia syndrome and/or lower cranial nerve palsies linked to long intubation due to COVID-19 ARDS were reported in the literature (Table 1) [12, 17].

All cases reported were male. Relationship between sex and cranial nerve palsies could be explained by the morphology of the male hyoid bone that favorize the local compressive cranial nerve [18]. All cases except one had unilateral nerve palsies. The patient with bilateral Tapia syndrome was not associated with prone-position but long length of mechanical ventilation [15]. Decavel et al. reported 10 cases on 88 patients admitted in the post ICU with at least one cranial nerve palsy. 90 % had a hypoglossal nerve palsy associated with a deficit in another cranial nerve in 9 of these 10 patients [17]. Here, we report in our 2 cases, the first case of a woman with Tapia syndrome. All of our patients received long duration of mechanical ventilation and several sessions of prone position ventilation.

## Conclusion

Because mechanical ventilation on prone position is a cornerstone of the treatment of hypoxic COVID-19 patients and lateral flexion of the head could induce a compression on the lower cranial nerves and their collateral branches, the number of patients with Tapia syndrome may increase in the future. A better knowledge would optimize tracking of Tapia syndrome and a faster rehabilitation procedure would improve the symptoms healings.

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## Author's Contributions

All authors have contributed to the drafting and the critical revision for the article. The final version was approved by all authors.

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## Availability of Data and Materials

All supporting data and images are available at demand.

## Ethics Approval and Consent to Participate

Due to COVID-19 pandemic situation, the ethical comity of the hospital has given its consent for any data collection in COVID-19 patients.

## Informed Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

## Competing Interests

All authors declare no competing interests.

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