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Research Article

Tapia Syndrome: Clinical Presentation, Diagnosis and Management of 40 Patients During Covid-19 Pandemic

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Abstract

Tapia syndrome is a rare combination of Vagus (X) and Hypoglosssal (XII) nerve palsy. It is a complication of prolonged intubation or airway manipulation of patients under general anesthesia with muscle relaxants. Incidence of this syndrome was observed to be increased in COVID-19 patients due to prone positioning and extended ventilation.

Objective: To analyze 40 cases of Tapia syndrome in COVID-19 patients, focusing on clinical features, risk factors and outcomes.

Methods: A retrospective review of 40 intubated COVID-19 patients diagnosed with Tapia syndrome was done. Data on symptoms, intubation duration, management, and recovery were analyzed.

Results: Patients presented with dysphonia, dysarthria and tongue deviation post-extubation. Mean intubation duration was 2 weeks. Treatment included corticosteroids, swallowing therapy and speech therapy. Those who did not recover with conservative management, surgical intervention with Autologous Fat Injection Laryngoplasty (AFI) was done. Overall 75.3% of patients recovered completely over a period of 6 months to 1 year.

Conclusion: Tapia syndrome is a notable complication in COVID-19 patients. Early diagnosis and supportive care improve outcomes. Also one should keep in mind that prolonged intubation in a prone position, change in head position with airway manipulation, throat packs, instrumentation in airway with head flexed are risk factors for development of this syndrome.

Keywords: Tapia Syndrome; COVID-19; Unilateral Vocal Cord Paralysis (UVCP)

Introduction

This case series presents an analysis of 40 patients diagnosed with unilateral vocal cord paralysis (UVCP) and Tongue deviation following a history of prolonged intubation during the COVID-19 pandemic. The study also explores the occurrence of Tapia Syndrome in a subset of patients and evaluates the outcomes of speech & swallowing therapy and surgical interventions.

Tapia syndrome overview

Tapia Syndrome is a rare neurological condition characterized by synchronous paresis or paralysis of the vagus nerve (CN X) and hypoglossal nerve (CN XII), commonly following prolonged intubation or airway manipulation [1,2]. It presents with dysphonia, dysarthria, dysphagia, and tongue deviation. The condition is believed to result from nerve compression due to flexion of the head and prolonged mechanical ventilation [3].

Mechanisms of injury include:

- Compression of the lower cranial nerves against stiff structures (cervical vertebrae, thyroid cartilage, or hyoid bone).
- Stretch injury due to overinflation of the endotracheal tube cuff.
- Beach chair positioning during surgery, leading to increased nerve vulnerability [5,6].

Previous studies have reported that recovery from Tapia Syndrome varies, with 30% achieving full recovery, 39% experiencing partial recovery, and 26% showing no improvement [7]. In this case series we present 40 patients diagnosed to have Tapia syndrome with hoarseness of voice, dysarthria, Tongue deviation, restricted tongue movements and aspiration. All patients had history of orotracheal intubation for a period of 1-2 weeks during the covid-19 pandemic and were referred to our department. This case series describes the etiopathogenesis, clinical features, investigation for prompt diagnosis and management strategy to manage this condition. Tapia syndrome can have severe morbidity and in some cases it is not reversible. Owing to the iatrogenic nature of injury, the awareness about this condition is very important.

Methodology

40 patients were retrospectively included in the case series who were referred to our OPD for evaluation of dysphonia and dysphagia. All patients had a history of intensive care unit admission and intubation for acute respiratory distress syndrome (ARDS). Average duration of intubation was 2 weeks. Post awakening patients had complaints of hoarseness of voice, dysarthria, tongue deviation, dysphagia, and cough bouts during meals. Duration of presenting symptoms ranged from 6 months - 1 year from the onset of symptoms. A thorough ENT examination was done. All patients were subjected to Videolaryngostroboscopy (VLS), Contrast-Enhanced Computed Tomography (CECT). VLS confirmed vocal cord palsy. Out of 40, 25 patients had Hoarsness of voice and Dysarthria.The remaining 15 patients had Hoarse voice, Dysarthria and Dysphagia. On CECT, no evidence of lymphadenopathy or mass compressing the nerve was seen.A neurologist opinion was sorted for all patients to rule out other central causes of Xth and Xth cranial nerve palsy. Patients with dysphagia, Functional endoscopic evaluation of swallowing (FEES) was done and then Ryles tube insertion was done and Swallowing therapy was initiated. All patients were started on corticosteriods therapy in the form of oral prednisolone at a tapering dosage of 1 mg/kg for 3 weeks. Supportive care treatment with Multivitamins and Speech therapy and Swallowing therapy was initiated. On follow-up visits after 1 month, 18 patients (45%) showed spontaneous recovery with good phonation and swallowing functions. Out of these 18 patients, 12 patients had Hoarse voice and Dysarthria and 6 patients had Dysphagia. 6 patients with Dysphagia follow up FEES was done which showed improvement and their RT removal was done and swallowing therapy was continued for 2 weeks. Oral corticosteroid therapy helped improve the symptoms in this group. 22 patients (55%) had persistent symptoms of hoarse voice and aspiration. These patients were then managed surgically by medialization Injection Laryngoplasty using autologous fat.

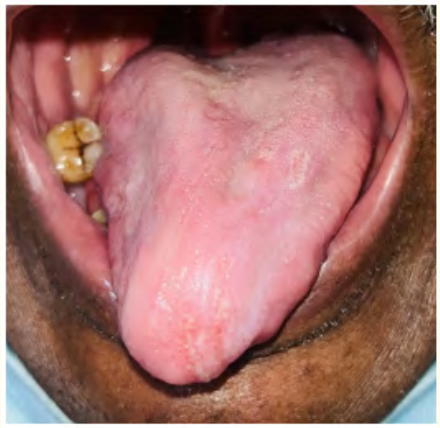

Image 1: Deviation of Tongue to the right side with atrophy of the right lateral border of tongue.

Surgical intervention (Autologous Fat Injection Laryngoplasty - AFI)

Performed in 22 patients. Out of these 22, 13 patients presented with symptoms of Hoarse voice and Dysarthria and 9 patients had symptoms of Dysphagia and aspiration also. Procedure was done under general anesthesia. Fat was harvested from the paraumbilical region, processed, and injected using an 18-gauge Brünings needle. Adequate approximation of the paralyzed vocal cord was achieved.

Follow up

Follow-up was done at 1 month, 6 months and 1 year post procedure. Out of 22 patients who were surgically managed with

AFI, 17 patients (77.3%) showed adequate glottic closure and resolution of initial presenting symptoms. 5 patients (22.7%) had residual symptoms of dysphonia and dysphagia. 2 patients (9.1%) had persistent phonatory gaps and hoarseness. 3 patients (13.6%) experienced fat absorption with persistent phonatory gaps and hoarseness. These 5 patients with persistent symptoms were offered revision surgery and continued voice therapy.

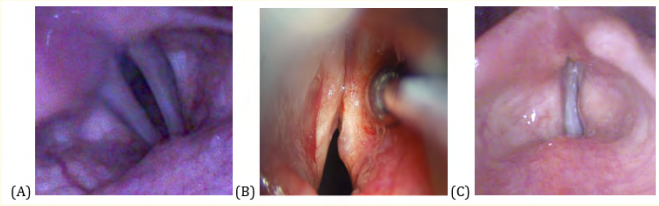

Image 2: (A) - Preoperative VLS of Right vocal cord palsy with large phonatory gap. (B) - Intraoperative image of Right vocal cord AFI. (C) - Postoperative VLS with adequate glottic approximation.

Results and statistical analysis

- Results analysed using Chi-square test
- A significant difference was observed between the spontaneous recovery group (45%) and those requiring surgical intervention (55%) with $p < 0.05$
- The success rate of AFI was 77.3%, with a failure rate of 22.7% (persistent symptoms or fat extrusion).
- Patients with Tapia Syndrome (Xth + XIIth nerve palsy) had a higher likelihood of requiring surgical intervention compared to those with isolated UVCP (Xth nerve palsy) ($p = 0.02$).
- Overall recovery rate of Tapia syndrome was 87.5% over a period of 6 months to 1 year from the period of initiation of symptoms.

Discussion

Tapia syndrome was first described in 1905 by a Spanish otolaryngologist, Antonio Garcia Tapia. He described this syndrome in a patient with unilateral vocal cord palsy and unilateral tongue deviation following a bullfighting injury with bull's horn pierced in the upper neck [1]. Subsequently patients with Xth nerve and XIIth nerve palsy were termed Tapia syndrome. Bilateral combination of nerve involvement is quite rare and is called B/L Tapia syndrome [2]. Both nerves originate from the skull base and they cross over in front of the transverse process of atlas. At this point both nerves are vulnerable to compression of stretching along the transverse process [3]. Both nerves lie along the lateral wall of the lower part of the tongue base and the oropharynx and upper part of the hypopharynx [3,4]. On the basis of duration of the injury, the nerve function may be temporarily impaired due to stretching (neuropraxia) or the nerve may be permanently damaged (neurotmesis) [4]. Mechanism by which the nerves may get compromised are hyperextension of the neck during intubation, flexion of neck, prone position, compression of nerve between endotracheal tube and transverse process of atlas, prolonged intubation, cuff overinflation, airway manipulation by instrumentation or tight throat packing [5]. In our study, 40 patients exhibited symptoms of Tapia Syndrome, demonstrating that prolonged intubation can lead to multiple cranial nerve dysfunctions.

Boisseau., *et al*. and Lykoudis., *et al*. [6] emphasized the role of head positioning and mechanical factors in nerve injury. Our results support these findings, as all patients with nerve involvement had a history of prolonged intubation and head flexion. Unlike these studies, our cohort did not have any structural abnormalities on imaging, reinforcing the theory of neuropraxia as the primary mechanism.

Gevorgyan and Nedzelski [7] reported a 30% complete recovery rate in Tapia Syndrome patients, which is comparable to our spontaneous recovery rate of 45%. This higher percentage in our study may be attributed to early initiation of speech and swallowing therapy. However, their reported rate of incomplete recovery (39%) and persistent deficits (26%) aligns with our findings, where 5 patients (22%) had persistent symptoms despite surgical intervention.

Wei and De Jesus [8] recommended MRI as a superior diagnostic modality to CT in ruling out central causes. However, our reliance on VLS and CECT was sufficient to rule out mass effects or other structural causes, reducing the need for further imaging.

Kraus., *et al*. [10] discussed the anatomical vulnerability of the recurrent laryngeal and hypoglossal nerves, suggesting that nerve compression between the endotracheal tube and surrounding

structures is a plausible mechanism. Our study reinforces this concept, particularly given the absence of any external compressive lesions in all patients.

The success of AFI in our study (77.3%) is in line with previous reports on fat injection laryngoplasty as a viable treatment for UVCP [6,7]. However, the 22.7% failure rate, including fat extrusion and persistent phonatory gap, highlights the need for refinement in technique and potential alternative interventions like Type I Thyroplasty or Laryngeal framework surgery [8,9]. Role of Laryngeal Electromyography and nerve reinnervation techniques can also be helpful. EMG plays a crucial diagnostic and prognostic role in Tapia syndrome, while nerve reinnervation strategies focus on optimizing functional recovery of the tongue and larynx [7,9-11].

Conclusion

This case series underscores the need for vigilance in post-intubation patients, particularly those with prolonged ventilation. Early diagnosis with VLS and CECT, followed by structured speech and swallowing therapy, can facilitate recovery. AFI serves as a valuable intervention for patients with persistent symptoms, though long-term follow-up is essential to assess outcomes and potential need for revision procedures.

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