Case Report

Tapia’s syndrome in post-operative patient following orotracheal intubation

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ABSTRACT

Introduction: Tapia’s syndrome is a rare condition that manifest due to unilateral extracranial nerve extension of cranial nerve 10 and 12 which occurred as a rare complication of Orotracheal Intubation in patient undergoing Laparoscopic appendectomy.

Case presentation: A 30 year old male, a known case of normal variant Right bundle branch block and Gilbert syndrome underwent orotracheal intubation prior to general anesthesia for emergency laparoscopic appendectomy. Postoperatively while assessing the patient there was deviation of tongue on left side.

Clinical findings and investigations: On examination of throat, atrophic and deviated uvula toward the right side was found. While protruding his tongue, tongue was deviated towards left side. Neurological examination revealed sluggish Gag Reflex. Brain Ct was done, which showed normal scan.

Interventions and outcome: Tablet Prednisolone and logopedic therapy in combination helped in early recovery; which is almost 8 weeks. Patient was completely recovered in 2 month with gradual improvement of phonation, tongue tone and mobility.

Relevance and impact: The intent of this report is to show how important it is for anesthesia providers and surgeons to understand Tapia’s syndrome, its causes, and the fact that it can occur despite seemingly normal airway care and neck posture. We believe that by raising awareness of this uncommon issue, practitioners will be able to early identify this complication.

1. Introduction

1.1. Background

Tapia’s syndrome is a rare condition first described in 1904 by a Spanish otolaryngologist Antonio Garcia Tapia. It is a unilateral extracranial combination lesion affecting the hypoglossal nerve (CN XII) and the recurrent laryngeal branch of the vagal nerve (CN X) that has been documented after general anesthesia for a variety of surgical operations [1]. Causative elements are thought to be surgical intraoperative neck posture and airway care [2]. Ipsilateral motor paralysis of the tongue and vocal cords is a symptom of this illness. Patients frequently experience dysphonia, dysphagia, and trouble swallowing after surgery [1,3]. Tapia’s case report is fascinating because of the rarity of the cases documented as well as the importance of early rehabilitation [4]. Peripherial Tapia’s syndrome is caused by the extra cranial lesion at the point of crossing of cranial nerve IX and recurrent laryngeal nerve which spares soft palate and pharynx motility, as the pharyngeal branch emerges more cranially [5]. However due to traumatic Tapia’s syndrome, we present a case with Uvular injury showing atrophic and right sided deviation which has also been reported in many other articles.

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1.2. Guidelines: SCARE 2020 paper

This case report has been reported in line with the SCARE Criteria [11].

1.3. Patient information: Demographics and presentation

A 30 year old male, Doctor by profession, unmarried, hindu by religion was presented to the Emergency Department (ER) with chief complaints of high grade fever (maximum recorded temperature of 104.8° Fahrenheit (F)). It was associated with generalized abdominal pain of moderate intensity, nausea and dizziness since 2 days.

1.4. Past medical and surgical history

He is a known case of normal variant Right bundle branch block and Gilbert syndrome. He had no significant surgical history.

1.5. Family history

He has a family history of hypothyroidism in his mother and his father died of myocardial infarction at the age of 56. He is a medical doctor by profession.

1.6. Drug and allergy history

No h/o of long term medication and no known allergies till date.

1.7. Social history

Doesn’t consume alcohol, Non Smoker.

1.8. Clinical Findings

Patient was ill looking and his vital sign upon arrival were normal with body temperature of 101.5 F, heart rate of 85 beats per minute, respiratory rate of 17 times per minute, and blood pressure of 130/80 mm Hg. Per Abdominal examination revealed tenderness and rebound tenderness on Right iliac fossa. His higher mental functions, sensory system, and cranial nerve were intact.

1.9. Diagnostic assessment

His laboratory findings revealed leukocytosis and raised C-reactive protein (CRP). Ultrasonography showed tubular, non-compressible, aperistaltic, blind ended tubular structure of 10.3mm noted in the right lower quadrant arising from the base of caecum without any fluid collection suggestive of acute appendicitis.

Emergency laparoscopic appendectomy under general anesthesia was planned and conducted after 12 hours of admission. During the preoperative periods, the patient showed no limitations in mouth opening and a Mallampatti score of class I. Intubation was tried few times by medical intern followed by residents before the successful intubation. Intubation was done on supine position. During the procedure Endotracheal tube, laryngoscope was used. The surgery was completed in 30 mins with minimal blood loss. Immediately after the effect of anesthesia reversal and extubation patient developed hoarseness, disturbed speech, and dysphagia. On examination of throat, atrophic and deviated uvula toward the right side (Fig. 1) was found. When he was asked to protrude his tongue, it was deviated towards left side (Fig. 2). He had a normal bowel and bladder habit.

Neurological examination showed sluggish gag reflex and rightward deviation of tongue. Other cranial nerves were intact and mini-mental examination was normal.

1.10. Brain CT scan was normal

1.10.1. Diagnosis and intervention

Traumatic pressure neuropathy in the left side is suspected during tracheal intubation. And a treatment of corticosteroid initiated with an initial dose of 40mg Prednisolone daily with slowly tapering over 4 weeks. Along with that, logopedic therapy was initiated to limit the possibility of permanent functional deficits and quickly recover swallowing and phonation.

Fig. 1. The image shows atrophic uvula and right sided deviation.
1.10.2. Outcome

Patient was completely recovered in 2 month with gradual improvement of phonation, tongue tone and mobility.

1.10.3. Intervention settings and name of clinician

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Dr. Prakash Poudel Jaishi, Department of medicine, Bhakunde Hospital.

2. Discussion

Tapia syndrome consists of a group of symptoms which occurs as a result of reversible compression of cranial nerve X (recurrent laryngeal branch) and XII(extracranial part). Recurrent laryngeal nerve has motor innervation to all the intrinsic part of larynx except cricothyroid muscle (supplied by superior laryngeal nerve) and has sensory innervation to the larynx below vocal cords. Injury to this nerve leads to paralysis of vocal chords [6]. Likewise, after emerging from hypoglossal canal, the extracranial part of hypoglossal nerve begins and has a motor supply to all the intrinsic and extrinsic muscles of tongue except palatoglossus (supplied by vagus). Injury to this part of nerve leads to difficulty in speaking (aphasia) and swallowing (dysphagia). Also, deviation of tongue to the affected side when asked to protrude the tongue [7,8].

B S Schoenberg et al. in their article emphasized that the symptoms are mainly due to ipsilateral hemiplegia of larynx and tongue with relative sparing of soft palate [10]. Many studies report that it takes 6 months to completely recover [9].

Fig. 2. Slight left sided deviation of the tongue at rest.

Our patient being a medical doctor was able to notice change in his voice, disturbance while speaking and difficulties while swallowing immediately after surgery and took immediate medical attention. Extensive neurological examination not only revealed injury to 12th cranial nerve, but also 9th and 10 cranial nerves function was also impaired. Patient being medical doctor identified his condition earliest and the main reason for early recovery is early identification of the condition. Early identification of condition, Early logopedic therapy along with steroid reduced the duration of illness to 8 weeks in contrary with many other studies [9].
2.1. Take away lesson

Endotracheal intubation in the setting of respiratory support in the intensive care unit can result in Tapia’s syndrome. To reduce the likelihood of occurrence, preventive steps should be considered. It is critical to obtain a fast diagnosis so that treatment and rehabilitation can begin as soon as possible in order to have a positive long-term outcome. Since, the patient himself was a doctor, his early identification and proper rehabilitation helped in early recovery of the patient. The intent of this report is to show how important it is for anesthesia providers and surgeons to understand Tapia’s syndrome, its causes, and the fact that it can occur despite seemingly normal airway care and neck posture. We believe that by raising awareness of this uncommon issue, practitioners will be able to early identify this complication based on clinical signs and further imaging including the management strategies.

2.2. Informed consent

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

Ethical approval

None.

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This case report hasn’t been funded by any person or any institutions.

Author contribution

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Consent

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Guarantor

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Declaration of competing interest

There is no any conflicts of interest with this article.

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