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Case Report
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Tapia Syndrome Plus Following Intubation for Transverse Sinus Stenting for Idiopathic Intracranial Hypertension - A Case Report

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Abstract

Background: Tapia syndrome has been defined as extracranial palsy of recurrent laryngeal nerve and hypoglossal nerve leading to unilateral vocal cord and tongue palsy.

Case Report: We report a case of atypical Tapia syndrome with recurrent laryngeal nerve, hypoglossal nerve and additional unilateral palatal palsy after orotracheal intubation in a patient undergoing transverse sinus stenting for idiopathic intracranial hypertension.

Conclusion: Occurrence of Tapia syndrome plus after orotracheal intubation is rare and unpredictable. Timely clinical suspicion can help in avoiding unnecessary investigations.

Keywords: Tapia syndrome; Intubation; Sinus stenting; Idiopathic intracranial hypertension; Cranial nerve palsy

Introduction

Vago-hypoglossal syndrome or Tapia syndrome, results from extracranial paralysis of vagus and hypoglossal nerves. A rare entity, it was first proposed by Antonia Garcia Tapia, a Spanish otolaryngologist in 1904, in a bull fighter who had vagal and hypoglossal palsy following neck injury [1]. It classically presents as dysphonia, dysphagia and deviation of tongue. This eponym signifies ipsilateral weakness of larynx and tongue while sparing function of the soft palate [2]. What is also notable is its causal association with orotracheal intubation and unpredictability of occurrence.

Case Presentation

A 28-year-oldhomemaker with gradually progressive bilateral blurred vision was diagnosed as Idiopathic Intracranial Hypertension (IIH) with bilateral transverse sinus stenosis in December 2020. She had CSF opening pressure of 400 mm of water and did not have desirable improvement with medical management. She was planned for venous sinus manometry and stenting. Orotracheal intubation with laryngoscope with Macintosh blade size 3 was done, tube fixed at 22 mm mark and the cuff was inflated at normal pressure. The intubation lasted two and half hours without undue flexion or lateral rotation. Venous sinus manometry showed gradient of 13 mm and 12 mm across narrowing in right and left transverse sinus, respectively (Figure 1a). The right internal jugular vein access was taken and balloon angioplasty was done at the level of narrowing bilaterally. However, there was no significant improvement in the pressure gradient. Thereafter, a self-expanding stent (8 mm × 40 mm) was deployed across the narrowing in right transverse sinus. Pressure gradient post stenting showed significant improvement and good flow was achieved across the stent (Figure1b, 1c). After wearing off of anesthesia, patient complained of change in voice with nasal intonation and difficulty in swallowing. There was flattening of the right palatal arch with deviation of uvula to left side, deviation of tongue to right and ipsilateral weakness of the tongue (Figure 1d). No other cranial nerve or focal neurological deficits were observed. In view of the preceding stenting procedure, a brain imaging was performed to rule out medullary involvement. MRI brain and MR angiogram were normal. Right sided vocal cord palsy was confirmed by direct laryngoscopy. Patient was put on nasogastric feeds and was given injectable steroids for 5 days followed by oral steroids. MRI brain

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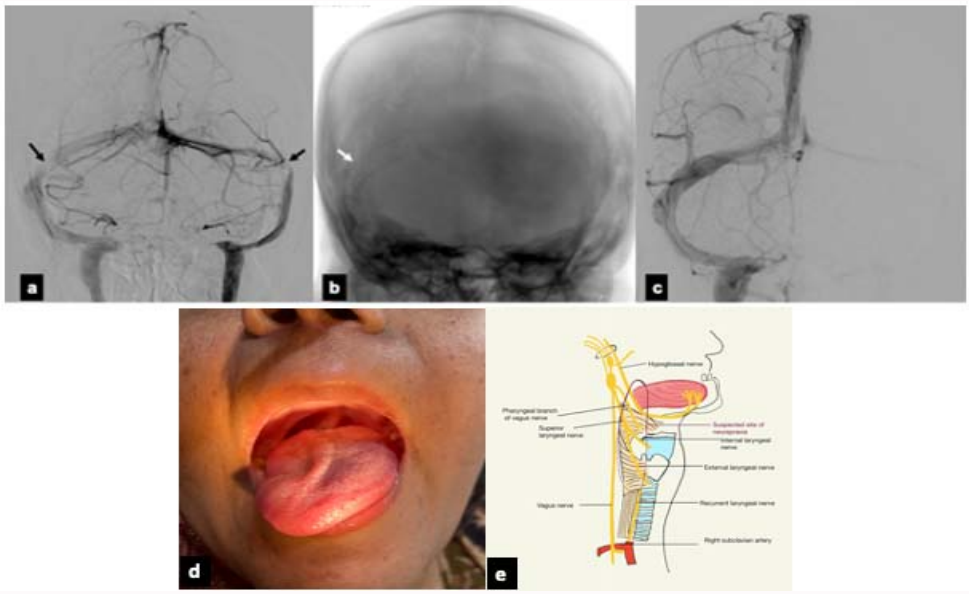

Figure 1: The venous phase of left vertebral artery injection (a) shows significant stenosis of bilateral transverse-sigmoid sinus (black arrows). Skull anterior-posterior axial view (b) shows the stent deployed across the right transverse-sigmoid sinus (white arrow). The post-stenting venogram (c) reveals good expansion of right transverse sinus. (d) Right vagus nerve palsy with deviation of uvula to the left and decreased palatal movement on the right side. Right hypoglossal nerve palsy with deviation of tongue to the right side. (e) Schematic diagram suggesting suspected site of neurapraxia through lateral laryngeal wall.

Table 1: Review of atypical presentation of Tapia Syndrome cases and structures involved in them.

S.No.	Author	Year	Age	Sex	Causative circumstances	Structures involved	Cranial nerves involved
1	Tapia et al. [2]	1904	31	M	Neck injury in bull fight	Unilateral tongue, Unilateral vocal cord, contralateral hemiparesis	Vagus nerve, Hypoglossal nerve, carotid artery contusion with cardioembolic stroke
2	Cariati et al. [3]	2016	42	M	Shoulder fracture open reduction	Unilateral tongue and vocal cord	Unilateral hypoglossal and recurrent laryngeal nerves. Lingual branch of trigeminal nerve
3	Boisseau et al. [4]	2002	42	M	Arthroscopy for recurrent shoulder dislocation	Unilateral tongue, vocal cord and soft palate	Unilateral hypoglossal nerve, recurrent laryngeal nerves and pharyngeal branch of vagus nerve
4	Sotiriou et al. [6]	2007	52	M	Coronary artery bypass grafting	Bilateral tongue and vocal cord	Bilateral hypoglossal and recurrent laryngeal nerves
5	Decavel et al. [7]	2020	62	M	Covid- 19 patient requiring mechanical support for 16 days	Unilateral tongue, vocal cord and soft palate	Unilateral hypoglossal nerve, recurrent laryngeal nerves and pharyngeal branch of vagus nerve
6	Turan et al. [8]	2012	15	M	Mechanical support in pneumonia	Unilateral vocal cord, bilateral tongue, loss of swallowing reflex for 24 hrs	Unilateral recurrent laryngeal nerve, bilateral hypoglossal nerve
7	Cinar et al. [9]	2005	20	M	Rhinoplasty	Bilateral tongue and vocal cord	Bilateral hypoglossal and recurrent laryngeal nerves

repeated after 10 days was unremarkable. At 30 days after onset, nasal intonation was persistent but patient was able to swallow without difficulty. At 6 months, patient had completely improved.

Discussion

Tapia syndrome has been noted following procedures like septorhinoplasty, shoulder surgery, cervical spine surgeries and with transient intubations where cardiorespiratory instability required intubation (Table 1) [1-7]. Bilateral Tapia syndrome has also been described following orotracheal intubation [8,9]. The proposed mechanisms following intubation include compression of nerves by pressure of the inflated laryngeal cuff across the lateral laryngeal wall and alternatively stretching of nerves due to undue head movements [1,5-7]. Tapia syndrome occurring after an intracranial procedure is unique due to the possible topographical association of intracranial lesions in relation to the procedure. Posterior fossa hemorrhage after stenting due to stent thrombosis has been noted and was ruled out in this case [10]. A similar presentation could be explained by the involvement of the nucleus ambiguous and hypoglossal nucleus or intramedullary fibers of the twelfth cranial nerve which was also excluded by neuroimaging. Another notable finding in our patient was the involvement of soft palate suggesting involvement of the pharyngeal nerve, branch of vagus nerve, which carries the motor impulses to the soft palate. Most of the described cases of Tapia syndrome involved hypoglossal and recurrent laryngeal branch of vagus nerve as compression is believed to occur at the crossing of these two nerves (Figure 1e and Table 1) [1-9]. Therefore, palatal involvement in our patient makes it a triad of pharyngeal, laryngeal and glossal involvement. One possible explanation for this can be the short neck length in our patient (midline neck length- 6 cm); making it more likely that neurapraxia also involved the pharyngeal branch exiting proximally (Figure 1e). This rarest entity can therefore be referred to as 'Triad of Tapia Plus syndrome'. Tapia syndrome has been found to resolve progressively in 6 to 12 months with steroids and dysphagia rehabilitation as in our patient [3,5].

Conclusion

Tapia syndrome, especially Tapia syndrome plus is a rare unpredictable situation occurring after orotracheal intubation. Through this case, the authors would like to emphasize the need for

high clinical suspicion by intensivists, neurologists and surgeons spanning all fields of medicine as timely clinical suspicion can help avoid unnecessary investigations.

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