Tapia’s syndrome: An unanticipated airway complication

Dear Sir,

Tapia’s syndrome is a rare clinical presentation characterized by neurologic deficits involving the hypoglossal nerve (XII) and a recurrent laryngeal branch of the vagal nerve (X).\[1\]

The first case of Tapia’s syndrome was reported in 1904 by otolaryngologist Antonia Garcia Tapia in a bullfighter who developed ipsilateral weakness of the larynx and tongue after a neck wound from a bull’s horn.\[2\] It can be of upper motor neuron (UMN) type or lower motor neuron (LMN) type. A lesion causes the UMN type in the nucleus ambiguous, the hypoglossal nerve’s nucleus, and the pyramidal tract. The LMN type results from concomitant injury to the nerves at the tongue base and the pyriform fossa, where the nerves lie nearby.\[3\] It occurs most commonly due to nerve injury but can also occur due to infection or tumor. Nerve injury can occur during airway manipulation, direct injury to the neck, prolonged intubation, during brachial plexus block, long abnormal neck position.\[4\] In our report, we would like to highlight that, apart from a complication of general anesthesia and intubation, extreme neck rotation to the opposite side by the interventional radiology team for internal jugular vein cannulation and cerebral angiography/stenting procedures, can also cause Tapia’s syndrome, which has not mentioned in the literature.

A 28-year-old female, weighing 50 kg, was admitted to the neurosurgery department in our institute with a headache and progressive decrease in vision for the last 7 months. There was no significant past medical history. She was diagnosed to have idiopathic intracranial hypertension. Her clinical and laboratory findings were unremarkable. She was planned for diagnostic cerebral angiogram ± venogram ± stenting under general anesthesia in Digital Subtraction Angiography (DSA) suite by interventional radiology. General anesthesia was induced with propofol (100 mg), fentanyl (100 micrograms), and muscle relaxation obtained with rocuronium (40 mg). A 7.5 mm endotracheal tube (ET) was passed easily (CL Grade-1) without any difficulty, and the cuff was inflated with air till there was no leak around the cuff. Anesthesia was maintained with isoflurane (1.2%), oxygen + air, and an intermittent bolus of fentanyl and rocuronium. After induction, the patient was handed over to the interventional radiological team. They performed the right internal jugular vein (IJV) cannulation under ultrasound guidance for venography and stenting. She underwent a diagnostic cerebral angiogram and venogram followed by stenting of the right transverse sinus for stenosis. The procedure lasted for 120 min. and was uneventful.

After the completion of the procedure, the patient was extubated and shifted to the recovery room. The patient had throat pain, mild hoarseness, and nausea in the recovery room, managed with analgesia and anti-emetics, respectively. On the next day, in the ward, the hoarseness and nasal intonation had increased in intensity, and she gradually developed difficulty in swallowing. Computed tomography brain was done immediately, which showed normal findings. Detailed ear, nose, throat examination showed hypoglossal nerve involvement (tongue deviated to the right side), and diagnostic nasal endoscopy examination showed right side vocal cord paralysis. MRI brain showed normal findings with no evidence of hemorrhage, ischemia, abscess. All laboratory and blood investigations were within normal limits. Neurology opinion was obtained because of bulbar weakness, and they started injection dexamethasone (4 mg IV 8 hourly). The patient improved symptomatically.

In our case report, we did not find any apparent cause of nerve damage. Several causes of Tapia’s syndrome, such as endotracheal intubation, fungal infection, vascular, traumatic, and neoplasm, are reported in the literature.\[4,5\] The reason for recurrent laryngeal nerve (RLN) paralysis is the compression of the anterior branch of the inferior laryngeal nerve in the posteromedial part of the thyroid cartilage. Most commonly left side recurrent laryngeal nerve gets paralyzed. Interestingly, our patient developed weakness of the right recurrent laryngeal nerve. The hypoglossal nerve can get injured if it is compressed by an ET tube or compressed between the laryngoscope and hyoid bone. It also can get compressed in excessive flexion of the neck [Figure 1].

One of the causes of Tapia’s syndrome, in our case, could be due to pressure from the laryngoscopy or a change in the position of the neck during intubation. Furthermore, it might also be attributed to the highly rotated head position made by the interventional radiologists during cerebral angiography and stenting.

Tapia syndrome, usually a diagnosis of exclusion, can be suspected with clinical history and examination of the neck...
and throat. CT and MRI of the brain and neck are essential to rule out the central cause. The recovery period for Tapia’s syndrome is between four and six months, depending on the extent of the lesion. The recovery of nerve function may be complete (30%), a partial recovery (26%), or incomplete recovery (40%). There is no established treatment; a short course of systemic steroids can be tried along with supportive treatment, including speech and swallowing therapy and steam inhalation.

Tapia syndrome can be prevented by careful positioning and gentle laryngoscopy during intubation, avoiding excessive pressure to the nerve during surgery, proper monitoring of cuff inflation pressure with manometer to avoid extreme nerve pressure, fixation of the tube to prevent unintentional movements of the inflated cuff cranially into the larynx, preventing extubation with inflated cuff and last, but not the least, careful positioning of throat pack to avoid excessive nerve pressure.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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