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CASE REPORT AESTHETIC

Tapia's Syndrome: An Unexpected but Real Complication of Rhinoplasty: Case Report and Literature Review

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Abstract Extracranial involvement of the recurrent laryngeal and hypoglossal nerves, also known as Tapia's syndrome, is a rare complication of airway management under general anesthesia. We report such a case after an otherwise uncomplicated rhinoplasty. After reviewing the other nine reported cases of Tapia's syndrome, we found that half of the patients had undergone rhinoplasty or septorhinoplasty. The risk factors, etiology, clinical course, recovery potential, and preventive measures are analyzed and discussed. Although Tapia's syndrome after rhinoplasty/septorhinoplasty appears to be rare, it should be considered by both the anesthetist and the plastic surgeon, and most importantly, special attention should be paid to preventive strategies.

Keywords Rhinoplasty · Complication · Tapia's syndrome

Tapia's syndrome, first described in 1904 by the Spanish otorhinolaryngologist G. A. Tapia [1], is due either to an intramedullary lesion of the nucleus ambiguus, the nucleus of the hypoglossal nerve, and the pyramidal tract (central type) or to extracranial involvement of the hypoglossal nerve and the recurrent laryngeal branch of the vagal nerve (peripheral type) [1, 2]. The peripheral type is characterized by neurological deficits involving both the hypoglossal and the recurrent laryngeal nerve as a result of concomitant injury to the aforementioned nerves at the base of the tongue and the pyriform fossa, where the nerves lie in very close proximity along their extracranial course. Several etiologies have been implicated, including oropharyngeal manipulations during anesthetic airway management [3–6]. The clinical signs, due to ipsilateral paralysis of the vocal cord and tongue, include hoarseness, uncontrolled tongue movements, disturbed speech and swallowing, and ipsilateral deviation of the tongue on protrusion and contralateral on rest [2–4]. Most of the cases are unilateral; however, bilateral involvement of the nerves has been reported [5, 7].

To date, nine cases of Tapia's syndrome attributed to manipulations during airway management for general anesthesia have been reported; interestingly, four of them occurred in patients who underwent rhinoplasty or septorhinoplasty. We report another case of Tapia's syndrome after rhinoplasty, along with a concise review of the pertinent literature.

The aim of this report is to draw plastic surgeons' and anesthesiologists' attention to this unexpected complication, mention the possible association of this rare complication with rhino/septorhinoplasty under general anesthesia, and summarize the potential preventive measures.

Anatomic Aspects

The hypoglossal nerve (CN12) is the motor nerve of the tongue. It leaves the skull through the hypoglossal canal in the occipital bone. It then enters the paravertebral space and descends almost vertically to a point corresponding with the angle of the mandible. It is at first deeply seated beneath the internal carotid artery and internal jugular vein, and then intimately connected with the vagus nerve. It then passes forward between the vein and artery deep to the posterior belly of the digastric muscle and reaches the submandibular region to enter the tongue. Before it enters the tongue, muscular branches of the nerve are distributed

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to the styloglossus, hyoglossus, and genioglossus. At the undersurface of the tongue numerous slender branches pass upward into the substance of the organ to supply its intrinsic muscles.

The vagus nerve (CN10) is composed of both motor and sensory fibers and has the most extensive course and distribution of any of the other cranial nerves. It passes vertically down the neck within the carotid sheath, lying between the internal jugular vein and internal carotid artery as far as the upper border of the thyroid cartilage, and then between the same vein and the common carotid artery to the root of the neck before it enters the thorax and then the abdomen. At the root of the neck it gives off the recurrent laryngeal nerve, which on the right side passes in front of the subclavian artery and on the left side passes to the left of the aortic arch. The recurrent laryngeal nerve on either side ascends in the groove between the trachea and esophagus, passes under the lower border of the constrictor pharyngis inferior, and enters the larynx. It is distributed to all the muscles of the larynx, except the cricothyroideus. Interestingly, damage to the vagus nerve before it gives off the recurrent laryngeal nerve can also cause paralysis of the recurrent laryngeal nerve. Concomitant paralysis of the hypoglossal and recurrent laryngeal nerves (Tapia's syndrome) can be caused by lesions affecting the lateral wall of the lower part of the oropharynx and the upper part of the hypopharynx where the hypoglossal and vagus nerves are in close proximity (Fig. 1).

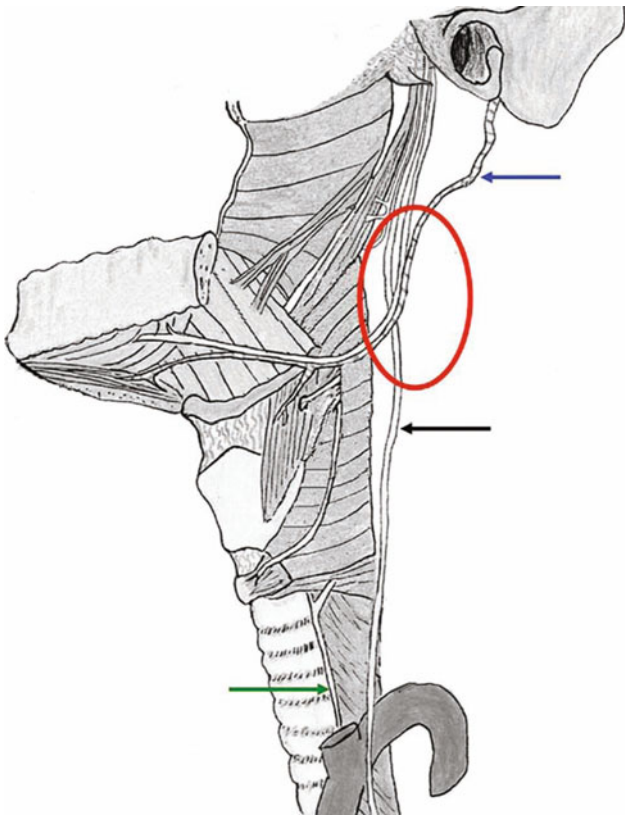

Fig. 1 Line drawing of the extracranial course of the hypoglossal (*blue arrow*), vagus (*black arrow*), and recurrent laryngeal (*green arrow*) nerves. In the *red circle* is the area of common course of the hypoglossal and vagus nerves (lateral wall of lower oropharynx and upper hypopharynx)

Case Report

A 32-year-old man underwent open rhinoplasty under general anesthesia. Endotracheal intubation and extubation were uneventful; the cuff was inflated to a pressure <20 cm H_2O and a throat pack was used. The first postoperative day the patient had all the physical signs of paralysis of the right hypoglossal and recurrent laryngeal branch of the vagal nerves. Therefore, the diagnosis of Tapia's syndrome was made since computed tomography of the head and neck was normal. Interestingly, the only finding on ENT examination was a slight contusion at the right side of hypopharynx, which was attributed to excessive pressure by the throat pack. The patient underwent conservative management with oral steroids together with speech and swallowing therapy. Hoarseness resolved 3 weeks postoperatively and hypoglossal paralysis resolved completely by 4 months.

Discussion

Injuries of several cranial nerves have been reported in the literature as a result of airway management for endotracheal general anesthesia. Nerve lesions and the resulting paralysis are attributed to stretching or compressing the nerves. The most commonly involved cranial nerves are the hypoglossal and the vagus. Concomitant paralysis of the hypoglossal and the recurrent laryngeal branch of the vagus, also known as peripheral-type Tapia's syndrome, has been reported in the literature.

In reviewing the literature, ten cases of Tapia's syndrome, including ours, attributed to manipulation during airway management for endotracheal general anesthesia, were identified. Interestingly, half of the cases occurred after endotracheal intubation for rhinoplasty or septorhinoplasty [3–6]. Yavuzer et al. [4] reported a case of unilateral Tapia's syndrome after septorhinoplasty, which was attributed to compression of the nerves after inflation of the cuff within the larynx. Cinar et al. [5] reported a case of bilateral Tapia's syndrome following rhinoplasty under general anesthesia. Although the authors state that there was no clear mechanism of nerve injury, a change in the position of the neck in combination with malposition or compression by the endotracheal tube was speculated.

Tesei et al. [6] reported another case of unilateral Tapia's syndrome after endotracheal intubation for rhinoplasty under general anesthesia. The lesion was presumably due to compression by the endotracheal tube and throat packing and/or neck hyperextension. No etiology was mentioned for the case of unilateral Tapia's syndrome reported by Boga and Aktas [3]. In our case, we speculate that the contusion noticed at the right side of hypopharynx was due to excessive pressure from throat packing that resulted in paralysis of the nerves.

The other 5 cases of Tapia's syndrome after endotracheal anesthesia reported in the literature occurred after cardiac surgery (2 cases) [7, 8], thoracic surgery (1 case) [9], shoulder surgery (1 case) [10], and repair of a fractured mandible (1 case) [11]. The etiology was also nerve injury due to stretching or compression as a result of mechanisms similar to those associated with rhinoplasty or septorhinoplasty. Although prolonged anesthesia may also aggravate the effect of the aforementioned manipulations, the mean operating time was less than 2 h in all the reported cases.

In regard to rhinoplasty/septorhinoplasty under endotracheal anesthesia, although any of the aforementioned causes may inflict nerve injury, we believe that throat packing plays a critical role, since (1) a throat pack was definitely used in four of the five cases of Tapia's syndrome (in the case reported by Boga and Aktas [3], it is not known whether a pack was used); (2) a throat pack is routinely used in rhinoplasty/septorhinoplasty cases; (3) a throat pack is applied in the hypopharynx and lower oropharynx, where both nerves lie superficially and in close anatomic proximity; and (4) throat packing also facilitates compression of the nerves upon the endotracheal tube and pharyngeal wall. Depending on the extent of the lesion, nerve function may be temporarily impaired (neurapraxia) with full recovery, which was the case in four patients, including ours, or permanently damaged, which was the case in one patient with partial recovery. Although there is no established treatment, supportive management by means of a short course of systemic steroids and vitamin B complex was practiced.

Interestingly, there are no reported cases of Tapia's syndrome after rhinoplasty or septorhinoplasty when general laryngeal mask anesthesia or sedation with local anesthesia is used; both of these types of anesthesia are commonly used during this surgery. When rhinoplasty/septorhinoplasty is performed under endotracheal anesthesia, both anesthesiologists and plastic surgeons should be aware of the following preventive measures suggested in the existing literature [3–11]:

1. Careful head and neck maneuvers during intubation, surgery, and extubation to avoid nerve stretching.
2. Gentle intubation to avoid direct trauma or excessive pressure to the nerves with the laryngoscope.
3. Careful extubation to avoid direct trauma or excessive nerve pressure.
4. Proper cuff inflation to avoid excessive nerve pressure.
5. Proper tube fixation to avoid inadvertent movements of the inflated cuff or extubation with inflated cuff, which could result in direct trauma or excessive nerve pressure.
6. Careful positioning of the proper amount of throat packing to avoid excessive nerve pressure.

Conflict of Interest The authors have no commercial interest or financial/material support in this study to declare.

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