

# Unilateral laryngeal and hypoglossal paralysis (Tapia's syndrome) following rhinoplasty in general anaesthesia: case report and review of the literature

## *Paralisi laringea e linguale unilaterale (Sindrome di Tapia) dopo intervento di rinoplastica in anestesia generale: caso clinico e revisione della letteratura*

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### Key words

Nasal surgery • Complications • Cranial nerve paralysis • Tapia's syndrome

### Parole chiave

*Chirurgia del naso • Complicanze • Paralisi dei nervi cranici • Sindrome di Tapia*

### Summary

Extracranial involvement of the recurrent laryngeal nerve and the hypoglossal nerve is known as Tapia's syndrome. Ipsilateral paralysis of the vocal cord and tongue is present. Lesion of these nerves may be a rare complication of airway management. Herein, a case of Tapia's Syndrome complicating transoral intubation during general anaesthesia in a rhinoplasty operation, together with a review of pertinent literature to evaluate the incidence and the possible pathogenic mechanism of the lesion.

There are recent reports in the literature on mono or bilateral paralysis of the XII or laryngeal recurrent nerve after use of laryngeal mask with a pathogenic mechanism of compression. Furthermore, there are reports, following oro-tracheal intubation, of recurrent laryngeal paralysis, likely legacies to the compression of the anterior branch of inferior laryngeal nerve by the cuff of the oro-tracheal tube against the postero-medial part of the thyroid cartilage. Hypoglossal nerve damage could be caused by a stretching of the nerve against the greater horn of the hyoid bone by a laryngeal mask or oro-tracheal tube or compression of the posterior part of the laryngoscope or oro-tracheal tube. In our case, the lesion probably occurred as the result of a two-fold compressive mechanism: on one hand, compression by the cuff of the endo-tracheal tube due to excessive throat pack in the oro-pharynx; on the other hand a prolonged stretching mechanism of these nerves may have occurred due to excessive anterior and lateral flexion of the head.

From the data reported in the literature, as in our case, complete recovery of function is generally achieved within the first six months. This progressive recovery of function suggests nerve damage of a neuro-praxic type, which is typical of compression injury.

In conclusion, the response of this rare complication confirms the importance not only of the position of the head and patient on the operating table but also the meticulous and correct performance of the routine manoeuvres of airway management.

### Riassunto

*L'interessamento extracranico del nervo laringeo ricorrente e dell'ipoglosso è conosciuto come Sindrome di Tapia, che consiste in una paralisi omolaterale della corda vocale e della lingua. La lesione di questi due nervi, può essere una rara complicazione delle manovre di gestione delle vie aeree. Riportiamo un caso di comparsa della Sindrome di Tapia come complicanza di una intubazione orotracheale in corso di anestesia generale per un intervento di rinoplastica. Si è provveduto alla revisione della letteratura per valutare l'incidenza e la possibile patogenesi della lesione. In letteratura vi sono segnalazioni recenti di paralisi mono- o bilaterali del XII nervo e del nervo ricorrente dopo l'uso di maschera laringea con un meccanismo patogenetico di compressione. Anche dopo intubazione orotracheale sono stati osservati casi di paralisi ricorrente verosimilmente legati alla compressione della branca anteriore del nervo laringeo inferiore esercitata dalla cuffia del tubo orotracheale contro il versante postero-mediale della cartilagine tiroidea. Il danno del nervo ipoglosso è invece riconducibile ad uno schiacciamento contro l'osso ioide da parte di una maschera laringea o ad una eccessiva compressione nella regione retrolinguale laterale ad opera di un laringoscopia o di un tubo orotracheale. Nel nostro caso l'ipotesi patogenetica è da ricondursi ad un duplice meccanismo di compressione: da un lato una compressione della cuffia del tubo endotracheale insieme ad una eccessiva pressione dello zaffo di garza introdotto in orofaringe; dall'altro è ipotizzabile un prolungato meccanismo di schiacciamento di questi due nervi dovuto ad una eccessiva flessione e rotazione laterale della testa. Dai dati della letteratura come nel nostro caso, generalmente il completo recupero della funzione si è osservata nei primi sei mesi. Questo progressivo recupero della funzione suggerisce un danno di tipo neuroprassico il che è tipico della lesione per compressione. In conclusione, il riscontro di questa rara complicanza testimonia l'importanza della posizione del paziente e della testa sul lettino operatorio e della meticolosa e corretta esecuzione delle manovre routinarie di gestione delle vie aeree.*

## Introduction

The extra-cranial involvement of the recurrent laryngeal nerve and the hypoglossal nerve is known as Tapia's Syndrome, first described, in 1904, by A. Garcia Tapia. Ipsilateral paralysis of the vocal cord and tongue occurs, with normal function of the soft palate<sup>1,2</sup>.

Injury of these nerves may be a rare complication of anaesthetic airway management<sup>3-7</sup>. Pressure neuropathy of both nerves, due to inflation of the cuff within the larynx, is an accepted cause. An alternative explanation is the stretching to both nerves<sup>8</sup>.

The present report refers not only to personal experience in a case of Tapia's Syndrome complicating transoral intubation for general anaesthesia in a rhinoplasty operation, but also a review of pertinent literature.

## Case report

A 30-year-old female (65 kg) underwent rhinoplasty under general anaesthesia, which was induced using a combination of a bolus of Remifentanyl (0.5 µg/kg) and Propofol 2 mg/kg. Muscle paralysis was obtained with Cisatracurium (0.15 µg/kg). Transoral intubation was performed with a Macintosh blade (No. 3) and a size 7 mm diameter reinforced tracheal tube was easily and gently placed in the trachea, on the first attempt and was then fixed to the right corner of the mouth.

No problems were encountered during laryngoscopy and intubation. The cuff of the tube was inflated with a pressure  $\leq 20$  cm H<sub>2</sub>O. No adjustment of cuff volume was made intra-operatively. N<sub>2</sub>O was not used for inhalation anaesthesia. The throat pack was placed in the pharynx to avoid the passage of blood in the aero-digestive tract.

General anaesthesia was maintained with Sevoflurane in oxygen/air and Remifentanyl in continuous infusion 0.25 µg/kg/min.

Moderate arterial hypotension was maintained with a cuff systolic artery pressure of approximately 90 mmHg. The operation was carried out in a semi-supine position with the head slightly inclined forward and laterally and trunk slightly elevated. Duration of the operation was 100 minutes. The patient was extubated, after removal of the throat pack without problems.

The following day the patient complained of difficulty in swallowing, dysphonia and hoarseness. Examination revealed deviation of the tongue to the right side and vocal cord paralysis, expression of hypoglossal and recurrent laryngeal nerve injury without local oedema or haematoma. The movements of the pharynx and soft palate were normal.

Meticulous neurological examination, including also magnetic resonance imaging (MRN) revealed no other evidence of central or cranial nerve involvement. Conservative management included steroids, vitamins together with speech and swallowing therapy. Full recovery of lingual and laryngeal functions was obtained within four months after surgery.

## Discussion

No surgical procedure is free of complications which may vary from common minor problems to very unexpected and severe episodes. In the case presented here, unilateral paralysis of the muscles of the tongue and ipsilateral vocal cord paralysis due to a lesion of 10<sup>th</sup> and 12<sup>th</sup> cranial nerves occurred following septorhinoplasty performed under oro-tracheal general anaesthesia.

This rare entity, known as Tapia's Syndrome, is believed to be caused by a neuropraxic reaction of both nerves due to pressure of the inflated cuff of the tracheal tube. To our knowledge, only four cases of unilateral Tapia's Syndrome have been reported following trans-oral intubation for general anaesthesia and only two following otolaryngologic procedures<sup>5,8-10</sup>. Yavuzer et al.<sup>10</sup> described one case of Tapia's Syndrome after septorhinoplasty that they believed to be caused by pressure neuropathy of nerves due to inflation of the cuff within the larynx. Boisseau et al.<sup>9</sup> reported a case following shoulder surgery in the sitting position. These Authors held that, in their patient, the compression by the tracheal tube, caused by displacement of the head, might have given rise to the nerve injury.

Hypoglossal nerve, lingual nerve and recurrent laryngeal nerve injury have recently been reported during the use of the laryngeal mask<sup>11-13</sup>. The precise aetiology of nerve damage associated with the laryngeal mask has not been determined, although erroneous positioning of the mask, excessive cuff pressure and position of the patient have been suggested as possible causes. It is also possible that the duration of the mask in the hypopharynx may be a factor.

Isolated unilateral or bilateral hypoglossal nerve damage, following transoral intubation, has been reported and the mechanism of injury is believed to be of neuropraxic origin due to pressure to the lateral roots of the tongue during routine intubation using a Macintosh blade with overextension of the head and the throat pack (tightly packed in the oro-pharynx)<sup>14-18</sup>.

Isolated unilateral or bilateral vocal cord paralysis have been reported following endotracheal intubation<sup>4,10,19</sup>. Compression of the anterior branch of the recurrent laryngeal nerve between the cuff of the displaced endotracheal tube and the posterior part of the thyroid cartilage was considered a likely mechanism<sup>4,10</sup>.

In the case of Tapia's Syndrome presented here, two probable mechanisms of nerve injury are considered: compression by the endo-tracheal tube upon the throat pack in the oropharynx, just at the point where the vagus and hypoglossal cross, that could have caused most of the findings in this patient such as difficulty in swallowing and hoarseness. Furthermore, a full-stretching mechanism of these nerves may have been due to excessive anterior and lateral flexion of the head. Anatomically, the hypoglossal nerve rests on the most lateral prominence of the anterior surface of the transverse process of C1<sup>20</sup> and crosses the vagal nerve. If hyperextension of this joint occurs, it is possible that these nerves would be stretched and

pressed against this prominence. To confirm this hypothesis, we have recently observed a case of Tapia's Syndrome secondary to direct injury of the soft palate and oro-pharynx by a foreign body.

The progressive recovery of the functions in this patient and in the majority of the cases reported in the literature suggest a neuropraxic type of nerve damage. However, logopedic therapy is often advisable. Although hypoglossal and recurrent nerve injury following routine endo-tracheal intubation appears to be rare, this complication should be considered both by the otolaryngologist and anaesthesiologist. To avoid such problems, special attention should be paid to correct positioning of the head during surgery.

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