



Case report

Bilateral palsy of the hypoglossal nerve following general anesthesia for emergency surgery. A case report



Panagiota Brattou^a, Konstantinos Iliakopoulos^b, Evangelos Anagnostou^c, Thomas Zambelis^c,
Andreas Polydorou^b, Kassiani Theodoraki^{a,*}

^a Department of Anesthesiology, Aretaieion University Hospital, National and Kapodistrian University of Athens, Greece

^b Department of Surgery, Aretaieion University Hospital, National and Kapodistrian University of Athens, Greece

^c Department of Neurology, Eginition University Hospital, National and Kapodistrian University of Athens, Greece

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ABSTRACT

Introduction and importance: Hypoglossal nerve palsy is a rare condition usually associated with tumors, trauma, stroke or multiple sclerosis. It can be associated with other cranial nerve palsies while injury to this nerve typically affects a patient's articulation by causing lingual motility disturbance and swallowing difficulty. Bilateral isolated hypoglossal nerve palsy is an even more infrequent condition, which can occasionally be due to airway manipulation.

Case presentation: We describe a case of bilateral hypoglossal nerve damage following general anesthesia for emergency surgery, presenting with dysarthria, immobility of the tongue and dysphagia after extubation. The patient had a gradual recovery of all lost functions during the next four months.

Clinical discussion: Bilateral hypoglossal nerve palsy is a very rare entity and tracheal tube malposition or prolonged but unnoticed tracheal cuff pressure especially in the face of low blood pressure, should be considered as possible causative mechanisms for this condition. This underlines the importance of careful positioning of the patient's head and neck during surgery as well as the meticulous and correct performance of routine maneuvers of airway management.

Conclusion: Bilateral hypoglossal nerve palsy is a very rare entity. Diagnosis and management of twelfth nerve palsy require a multidisciplinary approach to achieve the best patient outcome.

1. Introduction

The hypoglossal nerve (XIIth cranial nerve) provides the motor supply of the tongue and infrahyoid muscles. Palsies of the XIIth cranial nerve have been associated with tumors, stroke, head trauma, carotid endarterectomy, multiple sclerosis, Guillain-Barré syndrome and occasionally infection [1]. They usually tend to present unilaterally and are associated with the involvement of other cranial nerves and neural structures. Isolated hypoglossal nerve palsy has been described postoperatively with most reports presenting after orthopedic and otolaryngology procedures, while the majority of patients are male [2–5]. Most reports in the literature have a history of a surgical procedure under general anesthesia and orotracheal intubation, while inadvertent head misplacement under surgical drapes has also been implicated in some cases [2]. In few reports in literature, unilateral hypoglossal nerve

palsy has occurred after shoulder arthroscopic or open shoulder operations, where tilting the patient's head might have led to overpressure from the angle of the mandible to the hypoglossal nerve. In other shoulder surgeries, placing the patient intraoperatively to the beach-chair position might affect the posture of the head and neck, also causing unintended compression or traction on the hypoglossal nerve [2,3]. Dysphonia, dysphagia and ipsilateral tongue deviation towards the affected side upon protrusion on awakening from anesthesia are the classic presentation [4,5]. Hypoglossal nerve palsy is mainly diagnosed postoperatively after a thorough workup is performed to exclude other causes for the observed neurologic deficit, such as stroke, intracranial hematoma, impending airway obstruction, and endotracheal trauma [6]. Early consultation with otolaryngologists and neurologists is of paramount importance in guiding the diagnostic workup.

Bilateral isolated hypoglossal nerve palsy is an unusual entity with

* Corresponding author at: Department of Anesthesiology, Aretaieion University Hospital, National and Kapodistrian University of Athens, Vassilissis Sofias 76, 11528 Athens, Greece.

E-mail address: ktheodoraki@hotmail.com (K. Theodoraki).

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very few case reports in the literature [7,8]. Typical symptoms and signs are dysarthria, accumulation of saliva in the oropharynx making the patient to swallow frequently and dysphagia, due to the inability to properly direct food into the esophagus. Neurological examination reveals bilateral tongue atrophy and fasciculations, manifesting denervation-reinnervation injury [9]. In this case report, we present a case of bilateral hypoglossal nerve palsy in a patient after emergency abdominal surgery and highlight the importance of early consultation with otolaryngologists and neurologists to help in the identification of this serious condition.

2. Presentation of case

A 65-year-old male patient with a history of pancreatic cancer and without any other complicating illnesses was urgently readmitted to the operating room due to massive hemorrhage of the lower gastrointestinal tract and hypovolemic shock on the sixth postoperative day after a Whipple's surgical procedure. He suddenly became hemodynamically unstable with an altered level of consciousness, abdominal tenderness and anemia (Hb 7.5 g/dl).

Emergent administration of general anesthesia ensued and endotracheal intubation was accomplished with a video laryngoscope due to known difficult intubation from his previous surgery. Cricoid pressure was applied in the context of rapid sequence induction and an endotracheal tube of 8 mm internal diameter was inserted with relative difficulty with an anticlockwise maneuver and then was secured to the right angle of the mouth at 24 cm.

The patient continued to be hemodynamically unstable during

surgery and required vasopressors and transfusion of red blood cells and fresh frozen plasma. The operation was performed with the patient in the supine position and lasted 190 min. After surgery completion, he was transferred to the Intensive Care Unit, where he underwent mechanical ventilation for three days. Followingly, he was gradually weaned from ventilator support. After extubation, he presented with severe speech impairment, dysphagia, and inability to move and protrude his tongue. Pooling of saliva in the mouth made the patient swallow frequently. Otolaryngology review revealed fully mobile vocal cords and no significant edema present. On detailed neurological examination, pronounced dysarthria and severe speech impairment were present, while the patient's tongue was immobile, flaccid and rested on the floor of the mouth. There was normal sensation of the tongue, unaltered taste and no evidence of further cranial nerve involvement or other neurological deficits, while normal motility of the pharyngeal muscles and pharyngeal reflex was noted. The neurologists recommended a trial of pyridostigmine to improve dysphagia, which did not improve the situation. Laboratory tests, including serologic studies to exclude autoimmune disease or infection yielded normal results. A lumbar puncture for cerebrospinal fluid analysis was performed to exclude a possible malignancy with early manifestations, while radiography of the skull and the chest, computerized tomography of the head and magnetic resonance imaging of the brain and cervical spine ruled out intracranial compressive pathology or any central causes for the deficits observed (such as ischemic stroke or hemorrhage). Therefore, a diagnosis of bilateral isolated hypoglossal nerve paralysis was considered, which was confirmed by electromyographic examination that showed a complete bilateral denervation of the intrinsic tongue musculature (Fig. 1A). The

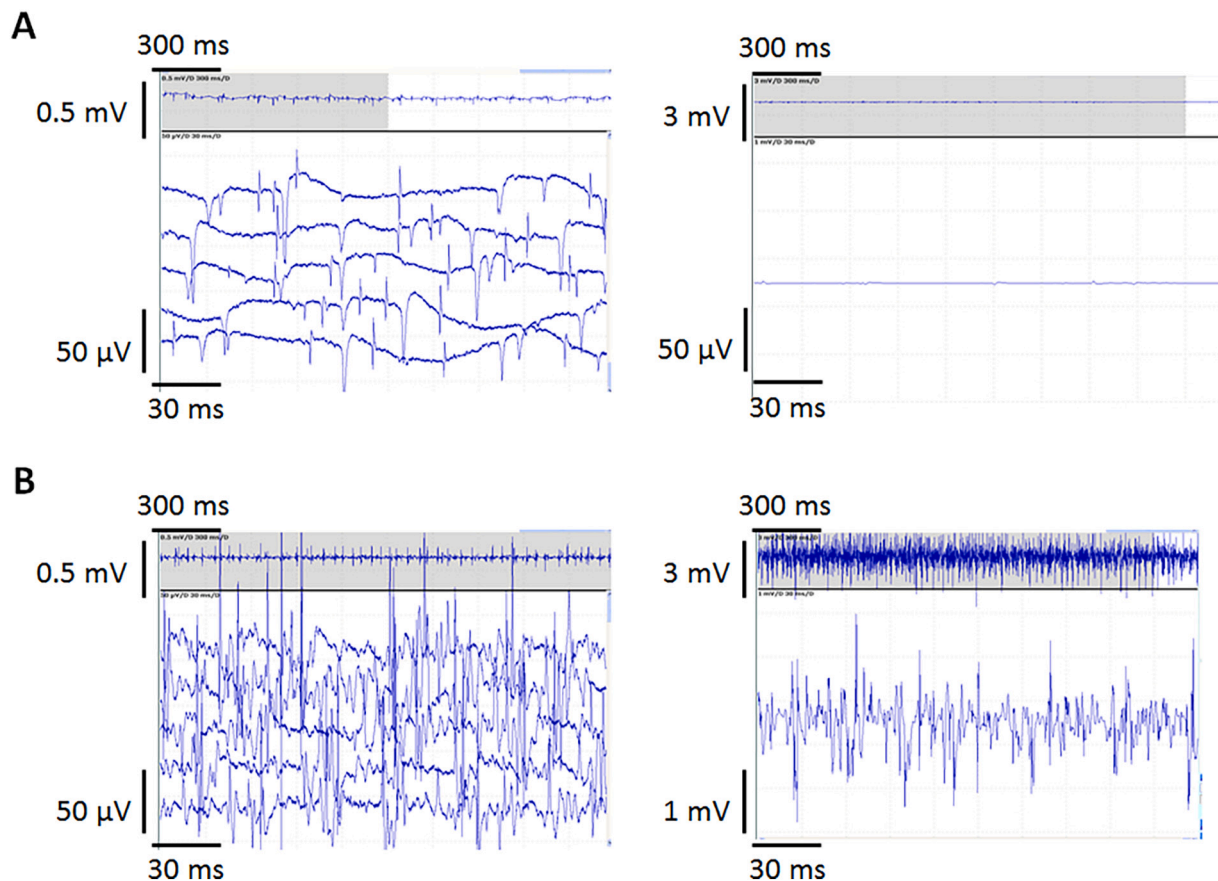


Fig. 1. A. Needle electromyographic recording of the intrinsic tongue muscles three weeks after extubation. At rest, abundant fibrillation potentials and positive sharp waves were observed, indicating acute tongue denervation (left). No voluntary activity could be produced, even at maximal effort (right). B. At follow-up four months after the previous recording, denervation activity at rest was not observed. The few potentials in the record are voluntary motor unit potentials arising from incomplete muscle relaxation (left). Voluntary tongue protrusion yielded a full interference recruitment pattern, designating normalization of tongue function (right).

neurologists recommended hydrocortisone 200 mg/24 h for four days without any significant short-term improvement.

During this period, the patient was dependent on a nasogastric tube for nutrition purposes and to prevent aspiration for a course of approximately 20 days, which unfortunately led to the prolongation of his hospital stay. The patient, of note, had spokesperson duties in his professional life beforehand, therefore psychological support was deemed necessary because the patient experienced the situation of impaired articulation as utterly debilitating and suffered severe anxiety. After discharge, he was placed on a soft food diet by which time the hypoglossal palsy had started to improve slowly. The patient was able to swallow his saliva satisfactorily as well as liquids and to extend his tongue symmetrically for a few centimeters one month later. He was able to swallow semi-solid food two months after surgery and eat normally three months after surgery. Four months after surgery, there were not any residual symptoms of dysphagia or dysarthria and the patient had returned to what was considered by him as "normality". At that time, complete recovery was confirmed by a follow-up electromyographic examination, which demonstrated a normal tongue motor unit recruitment pattern, without spontaneous denervation activity (Fig. 1B).

3. Discussion

Hypoglossal nerve palsy rarely occurs in isolation, while its occurrence postoperatively sparing other cranial nerves is very infrequent. To fully understand the mechanism of this incident, it is crucial to consider the anatomy of the nerve [9]. Anatomically, the hypoglossal nerve (cranial nerve XII) originates in the hypoglossal nucleus of the medulla. It passes between the olive and pyramid of the anterior brainstem before exiting the cranial cavity through the hypoglossal foramen of the occipital bone and after descending caudally, it travels ventral to the first cervical vertebra lateral mass and the facet joint of the first to the second cervical vertebra. The nerve lies approximately 2 to 3 mm lateral to the mid-anterior aspect of the lateral prominence of the first cervical vertebra [9,10]. Then, it receives contributions from cervical nerves as it enters the neck between the internal carotid artery and the internal jugular vein. Upon reaching the tongue's lateral aspect, it lies close to the hyoglossus muscle and inferior to the lingual nerve [1]. The hypoglossal nerve is exclusively a motor nerve and supplies all the intrinsic and extrinsic muscles of the tongue [9]. Therefore, considering the hypoglossal nerve's route, injury to the nerve can occur anywhere along its pathway, if it gets compressed or impinged between the endotracheal tube and stiff surrounding structures such as the cervical vertebrae and the thyroid and hyoid cartilage.

In most publications in literature, the common point is that all patients had oral intratracheal manipulation. Bilateral hypoglossal nerve palsy is very rare and the possible cause is usually related to either the endotracheal tube or its cuff and motion of the head during intubation. Tracheal tube malposition or excessive tracheal tube cuff pressure and/or volume or suboptimal airway management during laryngoscopy should be considered as possible causative mechanisms for this condition as they can lead to edema or injury to the underlying cranial nerves. As the hypoglossal nerve has a superficial course beneath the angle of the mandible, where it lies in close proximity to the tip of the greater cornu of the hyoid bone, nerve damage is also possible during mask ventilation. Based on the anatomical course of the hypoglossal nerve along the anterior surface of the transverse process of the first cervical vertebra, neck hyperextension during surgery in combination with the presence of a predisposition for calcification of the stylohyoid ligament can cause compression of the nerve against either the prominence of the cervical transverse process or the greater cornu of the hyoid bone [9]. Additionally, inadvertent extubation of the trachea with the cuff inflated, leading to impingement and stretching of the nerve against the greater horn of the hyoid bone, has been postulated as a cause for hypoglossal nerve injury.

In the case we describe, based on the history of known intubation

difficulty from the first operation, we may also have inadvertently exerted more pressure with the video laryngoscope blade both on the central aspect and on the lateral roots of the tongue to safely advance the tube and secure the airway quickly during the emergency procedure. This could have led to overstretching and shearing of distal nerve fibers of the hypoglossal nerves supplying motor input to the tongue or even soft tissue compression against the hyoid bone. With an inadvertent forceful maneuver during intubation and perhaps overzealous neck hyperextension due to the anticipated difficulty, the hypoglossal nerves may also have been impinged on the anterior aspect of the transverse processes of the first cervical vertebra. We also applied cricoid pressure due to the urgent nature of the reoperation, a fact which is also known to cause nerve compression against the hyoid bone or stretching of the hypoglossal nerves as they cross the hyoglossus muscle [9,11]. Finally, the endotracheal tube was railroaded with relative difficulty, as already mentioned, perhaps leading to direct compression of the hypoglossal nerves along their superficial course. Re-intubation has also been reported as a cause for hypoglossal nerve damage, as was the case in our patient, although the distance from the first to the second intubation (six days) makes this scenario a less likely cause for repeated iatrogenic trauma to the airway mucosa and underlying nerve structures [7]. However, our patient needed prolonged ventilatory support, which has also been reported as a cause for injury to the hypoglossal nerve and adjacent neurologic structures [12]. Finally, low blood pressure due to hemodynamic instability intraoperatively, the long operative time and perhaps sustained but unnoticed overinflation of the endotracheal tube cuff either high in the larynx or otherwise malpositioned, might have been a source of bilateral nerve ischemic injury and/or compression. These could be predisposing mechanisms in our case, perhaps in combination with the aforementioned factors.

In a large analysis of 69 patients with hypoglossal nerve palsy after airway management with general anesthesia, the majority of the patients were male [6]. The longer length of the greater cornu in men, the larger hyoid bone dimensions and the earlier ossification of the connection between the hyoid body and the greater cornu make compression at this level more possible as well as male patients more vulnerable to this injury [6,13].

As far as the treatment of this condition is concerned, management is largely supportive while rehabilitation has a leading role. Our patient did not want to take part in a speech and swallowing therapy relevant program, although this seems to be essential for a good outcome. The administration of steroids as part of the treatment armamentarium in the bilateral paralysis of the hypoglossal nerve remains controversial, without any clear evidence. Steroids are perhaps more helpful in case of airway edema and management should be guided by neurologists. Based on the disease's mechanism, early steroid treatment may reduce nerve swelling and promote recovery, given that there are no contraindications to steroid use. The present case demonstrated an excellent recovery without any residual neurological deficits despite the initial severe bilateral denervation of the tongue. Data from the literature show that hypoglossal nerve palsy appears to be largely self-limited. Forty-three % of patients had recovered by the first six weeks and another 40 % of the patients were symptom-free within six months of their operative date [6,7]. In our case, the gradual recovery of the deficits demonstrates a pattern of compression injury, compatible with neurapraxia rather than axonotmesis, which is characterized by much slower recovery or complete lack thereof.

The rationale for analyzing rare cases like this is to highlight the importance of identifying postoperative cranial nerve palsies and take proper measures to avoid similar incidences in the future. One might think that the use of intraoperative neuromonitoring for patients considered at higher risk is worth considering [14,15]. In fact, there have been reports of intraoperative hypoglossal nerve monitoring during brain and head and neck surgery, which showed some benefit. However, since this complication occurs so rarely, the routine utilization of intraoperative monitoring in cases not involving the head and neck

would likely not be cost-effective.

4. Conclusion

Isolated bilateral hypoglossal nerve palsy sparing any other nerve structures is a rare finding. However, awareness about this occurrence is necessary due to the associated significant morbidity related to its effect on swallowing and speech. It should be considered in patients after oral intubation for general anesthesia who report dysphagia, dysarthria, swallowing disability and palsy of the tongue. Diagnosis and treatment of bilateral paralysis of the hypoglossal nerve, as well as that of Tapia syndrome requires early multidisciplinary consultation to ensure patient safety. The collaboration of anesthesiologists, surgeons, radiologists, neurologists and otolaryngologists is crucial, while rehabilitation consists the mainstay of treatment. Psychological support is also necessary due to the severe emotional and mental trauma imposed on the patient until recovery. Due to the rarity of the syndrome, vigilance is required for early diagnosis and optimal patient outcome.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. The Institutional Review Board also provided consent for publication (436/06/06/2022). This work has been reported in line with the SCARE 2020 criteria [16].

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Consent

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Author contribution

PB: Conceptualization, Methodology, Project administration, Formal analysis, Writing-original draft. **KI:** Data curation, Investigation. **EA:** Data curation, Investigation. **TZ:** Data curation, Investigation. **AP:** Data curation, Investigation. **KT:** Conceptualization, Methodology, Project administration, Formal analysis, Writing—review and editing. All authors critically revised the manuscript for important intellectual content and approved the final version of the manuscript.

Research registration

N/A.

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Declaration of competing interest

The authors declare that they have no potential conflicts of interest.

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