Isolated hypoglossal nerve palsy post endoscopic micro-laryngeal surgery in a patient with systemic lupus erythematosus

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ABSTRACT
Hypoglossal nerve palsy (cranial nerve XII) commonly occurs in conjunction with other cranial nerve palsies. Isolated hypoglossal nerve paralysis is a rare complication of any procedures that do not involve dissection or incision of the neck. We report a case of a 42-year-old lady with underlying Systemic Lupus Erythematosus who developed isolated hypoglossal nerve palsy post-endoscopic microlaryngeal surgery for bilateral vocal cord polyps.

Keywords: Hypoglossal nerve palsy, microlaryngeal surgery, systemic lupus erythematosus

INTRODUCTION
Hypoglossal nerve palsy (cranial nerve XII) commonly occurs in conjunction with other cranial nerve palsies. Isolated hypoglossal nerve palsy is rare. They are generally caused by space occupying lesions of the internal or external cranium, metastatic tumours involving the base of skull, post-radiotherapy, oropharyngeal manipulation such as intubation and laryngeal mask airway (LMA), and to a much less common extent, connective tissue diseases such as systemic lupus erythematosus (SLE). Even more rare is the occurrence of isolated hypoglossal nerve palsies after procedures not involving dissection of incision of the neck.

CASE REPORT
A 42-year-old lady, with underlying SLE for the past 15 years, presented to the ENT clinic with hoarseness of five months duration. Her symptom was intermittent and was worsened with prolonged talking. There was no stridor, dysphagia, odynophagia or constitutional symptoms. There was no palpable cervical lymphadenopathy. All the cranial nerves were intact. On laryngeal examination, there were bilateral vocal cord polyps seen on the anterior one third of the vocal cords.

She proceeded to Endoscopic Laryngeal Microsurgery (ELMS) as a day-care pro-
procedure. She was intubated with a micro-laryngeal tube size 5.0 with prior muscle relaxation without any difficulties. The Benjamin-Lindholm laryngoscope was then inserted uneventfully. Cricoid pressure was applied throughout the ELMS procedure to achieve good visualisation of the vocal cords which lasted for about 10 minutes. The laryngoscope was then removed with no difficulties after the ELMS was completed. The patient was then extubated uneventfully. The total duration of anaesthesia was 30 minutes.

Four hours after the procedure, the patient complained of fever with chills and mild cough, and was admitted for observation. She was found to be febrile but other vital signs were stable. The total white cells were elevated with predominant neutrophils. Other parameters were within normal limits and the chest x-ray was normal. On post-operative day 1, she complained of difficulty with articulation and swallowing. She also complained of a left sided sore throat. On examination of the oral cavity, the tongue appeared deviated to the right with presence of fasciculations. Other cranial nerves functions were intact. A diagnosis of isolated right sided hypoglossal nerve paralysis was made. She was referred to Rheumatology for assessment in view of her underlying SLE that may have potentially caused the mononeuropathy. Her C-Reactive protein (CRP) was elevated (3.57 mg/dl) and Erythrocyte Sedimentation rate (ESR) was 56mm/hr. An MRI was performed to ascertain potential aetiologies. However no obvious pathologies were seen. The patient was given three 8mg doses of intravenous dexamethasone, followed by tapering doses of oral prednisolone. Swallowing assessment showed that there was good bolus manipulation with no intraoral residues post swallowing, and laryngeal elevation was appropriate with no signs of aspiration. She was then discharged on post-operative day 3. Her symptoms have improved and her tongue deviation was less marked at two months post-operatively. She is currently on a monthly follow-up.

DISCUSSION

The hypoglossal nerve is a pure motor nerve and supplies all the intrinsic and extrinsic muscles of the tongue except the palatoglossus muscle. The hypoglossal nerve is divided into six segments: supranuclear, medullary, cisternal, skull base, nasopharyngeal or oropharyngeal carotid space, and the hypoglossal segment. The hypoglossal nerve is at risk of being injured anywhere along its long course. Supranuclear lesions result in paralysis on the contralateral side of the tongue, causing deviation of the tongue towards the opposite side of the lesion. Lesion at the nuclear or infranuclear level leads to deviation of the tongue towards the side of the lesion, with associated atrophy of the intrinsic and extrinsic muscles with fasciculation. The findings in our case correlates with a lesion at the nuclear or infranuclear level.

Among the proposed theories in the development of perioperative hypoglossal nerve palsy include: oropharyngeal manipulation which results in stretching of the nerve against the greater horn of the hyoid bone by the use of an endotracheal tube (ETT) or a laryngeal mask airway (LMA), or compression of the nerve by the posterior part of the laryngoscope or ETT. Stretching of the nerve during intubation with cricoid pressure, and extu-
bation with the ETT cuff still inflated may contribute to the pathogenesis of hypoglossal nerve palsy.  

There have been many published cases of hypoglossal nerve palsy related to orotracheal intubation, with few reported cases linked to direct laryngoscopy for vocal cord lesions.  The most likely cause of the nerve palsy in our case was due to manipulation with the laryngoscope along with the cricoid pressure that was needed to be applied throughout the ELMS procedure. In our patient, it was unlikely that the nerve palsy was related to the underlying SLE, as the condition was not active and there were no other symptoms or signs that may suggest flaring or active SLE.

There have not been any recommendations on medical treatment for perioperative hypoglossal nerve injury previously. However, there have been theories that the use of steroid therapy may reduce oedema and potentially suppress the progression of the nerve paralysis. Even though hypoglossal nerve paralysis is a rare perioperative complication, we need to be more aware and cautious when handling with laryngoscopes.

In conclusion, we report a case of isolated hypoglossal nerve injury after undergoing an endoscopic micro-laryngeal surgery. Patients should be informed of this possibility, especially in complex cases.

REFERENCES
2: Cheng VS, Schulz MD. Unilateral hypoglossal nerve atrophy as a late complication of radiation therapy of head and neck carcinoma: a report of four cases and a review of the literature on peripheral and cranial nerve damages after radiation therapy. Cancer. 1975; 35:1537–44.