ORIGINAL RESEARCH PAPER

Volume-8 | Issue-2 | February-2019 | PRINT ISSN No 2277 - 8179

INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

# TRANSIENT ISOLATED UNILATERAL HYPOGLOSSAL NERVE PALSY ASSOSCIATED WITH HERPES SIMPLEX VIRUS- A CASE REPORT

Medicine

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## ABSTRACT

Transient isolated unilateral hypoglossal nerve palsy is rare diagnosis in clinical practice and various etiologies have been depicted in studies across the world. The sign and symptoms are milder than other palsies but it can be persistent or transient. There have been very few published single case reports on hypoglossal nerve involvement and case series are even fewer and we report a case of a transient isolated unilateral hypoglossal nerve palsy associated with Herpes Simplex Virus infection presented at SGT medical college, hospital and research center.

## KEYWORDS

Hypoglossal nerve palsy, Herpes Simplex Virus, Thornwaldt's cyst

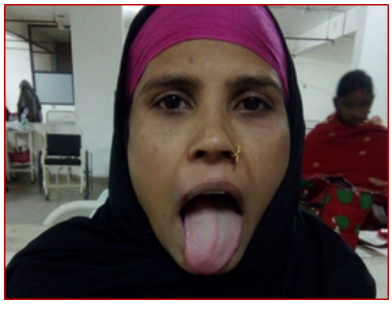
## INTRODUCTION

The hypoglossal nerve (12th Cranial Nerve) supplies the ipsilateral muscles of the tongue. The nucleus or the fibers of exit of hypoglossal nerve may get involved due to intramedullary lesions such as tumor, poliomyelitis or most often motor neuron disease. Involvement of cranial nerves outside the brain stem is frequently the result of trauma, localized infections including Varicella zoster virus and non-infectious (esp. carcinomatous) causes. [ 2]

This report describes the case of a 22-year-old female who presented with the deviation of tongue to the right side. Such deviation, atrophy and fasciculations of the tongue develop due to interruption of the 12th nerve. The differential diagnosis included neoplasia, trauma, infection, endocrine, autoimmune, neurologic, and vascular causes. Investigations included MRI, Chest X-Ray, NCV studies and a range of hematologic tests. It was a challenge to identify the etiology of this case

## CASE REPORT

A 22-year-old woman presented to the out-patient department with a history of dysarthria and dysphagia following a mild febrile illness from 2-3 days. She had deviation of tongue to the right side. (Figure 1) and had difficulty in chewing and swallowing. Taste was mildly altered.



**(Figure 1)**

She also had a complaint of pain in the right side of the occipital region of the head with mild swelling.

On general examination she had no signs of pallor icterus, cyanosis, clubbing, lymphadenopathy or edema. Her vitals were stable. However, she had herpetic lesions on the right side of the lip associated with burning sensation and pain radiating to the right side of the neck.

In Systemic examination respiratory cardiovascular, per abdomen was found to be normal. In CNS examination, she had tongue mounding and deviation to the right (Fig1)

All the facial muscles were normal. No sensory loss on the face. All other neurological and otorhinolaryngological findings were also normal. Patient did not have any hearing loss, ear fullness, sore throat, nasal discharge or any other nasopharyngeal sign and symptoms.

No history of tuberculosis, jaundice, seizures, diabetes, hypertension, drug intake or surgical illness was found. No significant history of headache, photophobia, vomiting or neck stiffness was found. Nausea was present. No altered level of consciousness, cranial neuropathies, hemiparesis or hemisensory loss was seen. No history of numbness or weakness was found.

## INVESTIGATIONS

Laboratorial investigations to rule out the causes of infections were carried out. Complete blood count, Liver function test, Kidney function test, Magnetic resonance imaging, Nerve conduction velocity, Rheumatoid factor, HIV1 & 2, VDRL, C-Reactive protein, Serology for HSV antibodies were done.

**The results are as follows**

**Complete blood count:** Hb: 11.3 gm/dl, TLC: 5,500/cmm, DLC: P50, L38, M02, E10, RBC count: 3.7mil/cmm, Platelet Count: 1.8L/Cm, MCV: 88.2fl, PCV: 33L%, MCHC: 34.2%, MCH: 30.2 pg. Absolute Eosinophilic Count: 450 cells/mr and ESR:10mm

**Liver Function Test:** Bilirubin: 0.3 mg/dl, Bilirubin Direct: 0.1 mg/dl, Bilirubin (Indirect): 0.2mg/dl, AST: 22.0 IU/L, ALT: 35.0 IU/L, ALP: 157.0 IU/L, Total Protein: 7.1 g/dl, S. Albumin: 3.8 g/dl, Globulin: 3.3H g/dL, A:G Ratio 1.1

**Kidney Function test:** Urea: 19.0 mg/dl, Creatinine: 0.88 mg/dl, Uric Acid: 3.9 mg/dl, Total Protein: 7.1 g/dl, Calcium: 7.9L mg/dl, Sodium: 136 mmol/L, Potassium: 4.4 mmol/L

**Urine examination:** Volume: 20ml, Color: Pale Yellow, Appearance: S. Turbid, PH: 6.0, Specific Gravity 1.025, Protein: NIL, Glucose: NIL, Red blood cells: NIL, White blood cells: 2-4 cell/HPF, Epithelial cells: 15-20 cell/HPF, Crystals: NIL, Casts: Granular positive, RBS: 95 mg/dl

**Special investigations** were also carried out:
Anti HCV antibody: Negative, HIV 1 and 2: Non-Reactive, VDRL/RPR: Non-Reactive, CRP (Latex): Negative, RA Factor (LAT): Negative, ANA: 0.17(Negative), ECG: WNL, NCV studies of upper and lower limb: Normal

MRI Brain was carried out which revealed a well-defined rounded lesion of size 9x9 mm(hyperintense on T2W and hypointense on T1Wsequence) seen in right paramedian location in the postero-superior aspect of nasopharynx s/o Thornwaldt's Cyst. It was an incidental finding in our case.

Antibodies for HSV 1 and 2, IgG and IgM were tested by Enzyme Immunoassay. The result was HSV 1 and 2 IgM was 0.32(Negative) whereas IgG was 3.47 (High). Titers of Herpes Simplex Virus and varicella-zoster virus antibodies were positive. Clinically also the

patient presented with the herpetic lesions on the right side of the upper lip on the upper lip. Hence, infection with Herpes Simplex Virus was confirmed.

**Follow up:** The patient was treated with Acyclovir 400 mg thrice daily and Cefixime 200 mg twice daily with multivitamins such a Methylcobalamine for 7 days. Her recovery was complete. [Fig. 2]

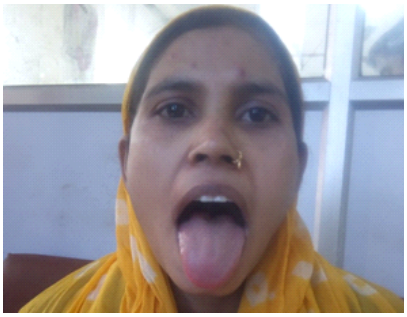


**Fig. 2**

## DISCUSSION

Our case is a young 22-year-old female who presented with transient isolated unilateral hypoglossal nerve palsy due to infection with Herpes Simplex Virus and the review of literature provides information about different etiologies, prognosis and management described below.

There is an interesting study by Kaene et al where 100 cases of 12th nerve palsy was studied to understand the etiology. The clinician had put 26 years of his experience and brought to our knowledge that half of the cases of Hypoglossal nerve palsy were found to be due to tumors (46 cases). One third of the cases were found to have Hypoglossal nerve palsy due to multiple sclerosis, stroke, surgery. Only four percent patients had Hypoglossal nerve palsy secondary to an infection. [3]

Nelson et al reported two case studies of progressive Hypoglossal nerve palsy where one of the patients was 58-year-old and presented with tongue weakness from 6 years. MRI and MR angiography revealed extensive paranasal pansinusitis and was previously operated for the same. Pansinusitis did not appear to affect the course of the hypoglossal nerve palsy and hence it was at lastly termed idiopathic. The other case reported by Nelson et al presented with viral upper respiratory illness which worsened the already existing Hypoglossal nerve palsy of two years duration. In both the cases Hypoglossal nerve palsy was progressive. [4]

Borden at al reported a similar case where 36-year-old man had a difficulty in moving the tongue and chewing. A week before, he developed a moderate to high intensity, right-sided hemi cranial headache that started in the cervical region, radiating to the occipital region of the head. A head magnetic resonance imaging (MRI) scan only revealed a cyst of the septum pellucidum. Otorhinolaryngology department examined the patient which did not identify any loco-regional cause that may explain the patient's symptoms. The patient recovered within 10 days. [5]

Ramzi et al reported two cases of Hypoglossal nerve palsy. One of them presented with reduced movement of the tongue and the second case presented with severe pharyngitis. Etiology was found to be secondary to infective mononucleosis. The possible explanation is presumed to be a local viral infection around the hypoglossal nerve nucleus. [6]

One of the largest case series of Hypoglossal nerve palsy describes the study of 12 patients by Sharma et al. The inclusion criteria were unilateral monosymptomatic Hypoglossal nerve palsy without any other neurological deficit such as bell's palsy, hemiparesis etc. They found that 50 % of the patients had Hypoglossal nerve palsy due to tubercular CNS infection. 25 % were found to be idiopathic and no etiology was figured out. One third of the cases were due to neoplastic, sarcoidosis, trauma, surgery etc. Two cases of Hypoglossal nerve palsy that died had neoplastic infiltration of the tongue and Chordoma respectively. Rheumatoid arthritis was also found to be the cause in one of these 12 patients. Most of the cases resolved and had potentially treatable etiologies. [9]

Shikino et al reported a case where a 33-year-old woman presented with a complaint of dysarthria and dysphagia from five days. Other neurological and otorhinolaryngological findings were normal. Titers of HSV and Varicella antibodies were found to be positive but could not differentiate between the acute and convalescent phase. MRI was found to be a normal. Idiopathic isolated unilateral hypoglossal nerve palsy was diagnosed secondary to HSV infection. Patient recovered without intervention. [7]

Not only adults but pediatric cases were also reported. In a case report by Yoon et al a 11-year-old Korean child was admitted with sudden onset of dysarthria, tongue fasciculations and weakness. All other investigations were normal except Epstein Barr virus nuclear antigen which was found to be positive, EBV VCA IgG was positive. All other findings including MRI was normal. Solumedrol was given for 12 days and complete recovery was seen. [8]

Its worth mentioning the case reports of Thornwaldt's cyst also as it was seen in was detected in our patient also. After extensive review of literature we would like to mention the study by Kumar et al where they reported a case of four patients with Thornwaldt's cyst. All of them were incidentally detected. The sign and symptoms of these patients were mostly nasopharyngeal such as one of them presented with pain on swallowing, nasal regurgitation of food and rhinolalia while the other presented with nasal obstruction, halitosis and headache. None of these patients presented with Hypoglossal nerve palsy. [10]

Another huge Thornwaldt's cyst was reported by Lin et al. They reported the case of 53 years old complaining of progressive nasal obstruction. Endoscopic examination revealed a 4cm large Thornwaldt's cyst. Hypoglossal nerve palsy was not seen. [11]

Baisakhiya et al reported a similar case of nasal obstruction for three months. Thronwaldt's cyst was confirmed in MRI report No tongue abnormality was seen. [13]

Hegde et al also reported a case of a patient who had a complaint of nasal obstruction for 10 years, headache hearing loss, ear fullness, otorrhea etc. MRI revealed Thornwaldt's cyst but Hypoglossal nerve palsy was not seen Hypoglossal nerve remained unaffected. [12]

To the best of our knowledge and review of literature, Hypoglossal nerve palsy was not seen in patients with Thornwaldt's cyst. Small cysts and usually asymptomatic and large ones cause nasal obstruction, halitosis, eustachian tube defect, ear fullness, occipital headache, post nasal drip, changes in olfaction and other nasopharyngeal symptoms.[12]

## SUMMARY

We report the case of transient isolated unilateral hypoglossal nerve palsy associated with Herpes Simplex Virus. It is stated that HSV is a double standard DNA Molecule. HSV 1 and HSV 2 are similar with genomic homology of 50%. It produces infections, which involve mucocutaneous lesions, CNS and sometimes visceral organs. Both virus subtypes can cause oral facial and genital infections, and clinically indistinguishable. Clinical signs and symptoms include fever malaise myalgia, inability to eat, irritability and cervical adenopathy and may last 3-14 days. Lesions may involve hard and soft palate, gingiva, tongue, lip and facial area.

HSV affects CNS and PNS also. In children and young adults primary HSV infection may result in encephalitis. Neurological sequalae of HSV is rare. HSV is the most commonly identified cause of recurrent lymphocytic meningitis. Autonomic nervous system is also affected in many cases leading to numbness and tingling, urinary retention, constipation, CSF pleocytosis, impotence may also occur. Hypoesthesia of the area of the skin innervated by trigeminal nerve and vestibular system dysfunction are the predominant signs of the disease. Rarely transverse myelitis, Gullian barre syndrome, Bell's palsy or cranial polyneuritis may be related to reinfection of HSV infection. [1] Our case is similar to the case report by Shikino et al as HSV titers were found to be high and patient had a recent infection of HSV.

Thornwaldt's cyst is defined as an inconstant blind sac located above the pharyngeal tonsil in the midline of the posterior wall of the nasopharynx. It's a developmental abnormality and is usually asymptomatic or an incidental finding. The sign and symptoms found in their case study included occipital headache, Post nasal drip, nasal

obstruction, nasal regurgitation etc. In the studies reviewed by us regarding the Thornwaldt's cyst, none of them presented with hypoglossal nerve palsy. Our case is also slightly similar to the study by Borden et al where they performed a head magnetic resonance imaging (MRI) scan which revealed a cyst of the septum pellucidum. Our patient revealed a Thornwaldt's cyst which is found to be benign and insignificant in size and did not affect the course of the hypoglossal nerve.

Also, it should be noted that the patient had developed the symptoms of hypoglossal nerve palsy soon after developing the lesions on the upper lip which is diagnosed as Herpes simplex virus infection.. So, in our opinion the cause of hypoglossal nerve palsy in this patient is an infection with Herpes Simplex Virus

## CONCLUSION

In this case of a 22-year-old female with deviation of the tongue to the right side along with herpetic lesion on the lip on the same side, detailed history, examination, and investigation were carried out to rule out the many causes of such a presentation. MRI revealed a Thornwaldt's cyst and the HSV serology revealed IgG antibodies positive for HSV. Most of the literature or case reports of Thornwaldt's cyst had nasal and pharyngeal sign and symptoms, where as a case of Hypoglossal nerve palsy following HSV infection is mentioned in the review of literature. With the best of our knowledge and the review of literature we conclude that HSV infection was the etiology of Hypoglossal nerve palsy in this case and the patient recovered with treatment within 4 to 6 weeks.

This case is reported for its rarity. Isolated hypoglossal nerve palsy is usually termed Idiopathic due to difficulty in concluding the etiology. In our case isolated unilateral hypoglossal nerve was associated with Herpes Simplex Virus infection and it was transient and not persistent.

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