

Case Report

A rare case of herpes zoster maxillaris with cranial polyneuropathy

Shubhransu Patro¹, Debananda Sahoo^{1*}, Anil Kumar Dash², Biswamohan Mishra¹

¹Department of Medicine, ²Department of Otolaryngology and Head and Neck Surgery, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India

Received: 19 September 2016

Accepted: 22 October 2016

*Correspondence:

Dr. Debananda Sahoo,

E-mail: drdebanandasahoo@gmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Varicella zoster virus has a high affinity for the ganglion and causes various neuropathies. About 12% of all peripheral facial nerve palsies are caused by varicella zoster virus, a rare complication due to reactivation of the herpes zoster virus in the geniculate ganglion of facial nerve. The occurrence rate of associated cranial polyneuropathy has been reported to be 1.8-3.2% and cranial nerves VII, VIII, IX and X are the ones most commonly affected. Here, we present a case of Herpes zoster maxillaris with cranial polyneuropathy who was improved with oral anti-viral drugs and steroid. Multiple cranial nerve palsies are being observed frequently in patients who were referred with a preliminary diagnosis of facial palsy. These patients are required to be examined in detail for involvement of other cranial nerves. Early diagnosis and treatment is required for reversing the functions of cranial nerves and decreasing the chance of developing other complications.

Keywords: Acyclovir, Geniculate ganglion, Herpes zoster maxillaris, Varicella zoster virus

INTRODUCTION

Herpes zoster virus involving the head and neck region usually presents with varied symptoms. Here we present a case of VZV infection with multiple unilateral cranial nerve palsies i.e. V, VII, IX, X and XII.

Primary infection to Varicella zoster virus (VZV), a DNA virus of the herpes viridae family, produces the typical picture of chickenpox. After primary infection occurs generally during childhood, the virus remains quiescent. The most frequent sites of latency of VZV are dorsal root ganglia of the spinal nerves, but any autonomic ganglion or cranial nerve can be involved. Reactivation of this virus during adulthood can produce zoster (shingles), which presents with painful vesicles with a dermatomal distribution.¹

Involvement of the ophthalmic division of the trigeminal nerve (herpes zoster ophthalmicus) and involvement of the facial nerve (herpes zoster oticus) are the two most

widely known clinical presentations regarding cranial nerves. Involvement of other cranial nerves by VZV is more rarely reported. We present a case of VZV infection of the left sided palate (maxillary division of cranial nerve V) with neuropathy of other four ipsilateral lower cranial nerves (VII, IX, X, and XII).

CASE REPORT

In May 2016, a 48 year old male was presented with chief complain of dragging of mouth to right side since last 5 days, associated with pain inside mouth, difficulty during movement of tongue, difficulty in deglutition with regurgitation of food through nostril, and pain and watering from left eye. There was no hearing loss or dizziness. There was associated dribbling of saliva from left side of mouth.

On physical examination, patient was in a stable clinical condition with intact sensorium. Multiple vesicular eruptions with ulcerations were present on the left side

palate (Figure 1). Palatal and Pharyngeal reflexes were absent on left side. There was infranuclear type of palsy of left 7th cranial nerve along with Bell's eye phenomenon (Figure 2). Tympanic membrane was intact. Extra ocular movements were intact and both the pupil were normal in size and reacted to light bilaterally. Tongue was deviated to right on protrusion (Figure 3). Also there was ipsilateral (left side) paralysis of the soft palate.



Figure 1: Multiple vesicular eruptions with ulcerations on the left side of hard palate.



Figure 2: Bell's eye phenomenon on left side.

Other systemic examination including the examination of neurological system revealed no abnormality except loss of taste sensation of anterior 2/3rd of tongue on left side. The patient was admitted and investigated. All blood reports like CBC, FBS, PPBS, LFT and RFT were within normal range. MRI scan of brain revealed no abnormality. The diagnosis was confirmed by detecting IgM antibody to varicella zoster virus in blood by ELISA method. CSF sample was positive by PCR and showed amplification of VZV DNA.



Figure 3: Tongue deviated to right on protrusion.

During hospital stay, the patient was treated with oral acyclovir 800 mg 5 times a day for 7 days and a short course of oral prednisolone 60 mg per day for 10 days along with other supportive medications and care like physiotherapy with an impression of multiple lower cranial nerve involvement by herpes zoster virus. After 3-4 days of admission, the mucosal lesions over the palate were healed but the paretic symptoms were persisted. On 10th day of admission, he was discharged. On revisit after 2 weeks, the mucosal lesions were completely healed up and left sided facial palsy was minimal with improvement of pharyngeal and palatal movements. Patient was advised for revisit after one month again. On subsequent follow up visits, patient improved a lot clinically with mild residual palsy of lower cranial nerves of left side and was advised to continue physiotherapy.

DISCUSSION

Facial nerve palsy is well known as Ramsay Hunt syndrome and is caused by reactivation of the Varicella zoster virus. Patients with Ramsay Hunt syndrome typically present with a peripheral facial nerve palsy associated with sensory neural hearing loss, tinnitus, vertigo with nystagmus and painful vesicular eruptions. However, numerous articles have recently reported various cranial neuropathies, other than those of the facial and acoustic nerves.²⁻⁵

After primary infection, the varicella viruses usually remain dormant in sensory dorsal root ganglia and activated from latency in elderly, diabetes and immunocompromised patients. Reactivation of varicella from geniculate ganglion causing LMN type of 7th cranial nerve palsy and vesicular rash in external ear along with severe otalgia comprises the Ramsay Hunt syndrome.

It rarely accompanies multiple cranial nerve involvement. Herpes zoster also involves the mucous membrane of the tongue, palate, pharynx, and larynx. Herpes zoster

infection of the larynx accompanied by RHS with cranial polyneuropathy is extremely rare, with only few reported cases in the literature.^{1,6-9} About 12% of all peripheral facial nerve palsies are caused by varicella zoster virus. The occurrence rate of associated cranial polyneuropathy has been reported to be 1.8-3.2% and cranial nerves VII, VIII, IX and X are the ones most commonly affected.¹⁰⁻¹²

In our case, the skin lesions were absent, but there was mucosal lesions confined to left side of the palate with associated LMN type of left sided facial palsy and involvement of ipsilateral IX, X and XII cranial nerves. Besides these, oropharyngeal discomfort and complaints such as dysphasia and pain inside mouth were more troublesome symptom due to paralysis of the left sided pharyngeal muscles and the palatal lesions.

Although MRI scan of brain can show the enhancement of the involved nerve in some cases, the findings do not provide information about severity or prognosis.^{13,14} In our case even with multiple cranial nerve involvement, no enhancement of involved cranial nerve (CN) was noted on MRI with gadolinium enhancement. The cause of multiple CN involvement has been explained as a consequence of an inflammation-induced infarction of a small vessel knowing that a small branch of the carotid artery supplies two or three contiguous nerves.¹⁵ Also the occurrence of anastomosis among 5th, 7th, 9th and 10th cranial nerve explains why the symptoms occur in these nerves. In addition, cranial nerves neighbourhood in the cavernous sinus also explain the theory of herpes zoster with multiple cranial nerve palsy.¹⁶ As the patient was able to take semisolid diet orally, he was given oral acyclovir along with oral steroid. There was no statistical significant difference in the outcome were noted among patients treated with oral and intra venous acyclovir. Recent data suggests that treatment with acyclovir prevents the permanent facial palsy.

ACKNOWLEDGEMENTS

Authors would like to acknowledge the Kalinga Institute of Medical Sciences authority for the support for managing the case.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

- Steiner I, Kennedy PG. Herpes simplex virus latent infection in the nervous system. *J Neurovirol* 1995;1(1):19-29.
- Kikuchi H, Yoshimura T, Hara H. A case of multiple cranial neuropathy due to varicella-zoster virus infection: detection on involvement of cranial ganglia with MRI. *Rinsho Shinkeigaku*. 1995;35:814-6.
- Nagano K, Yoshimura K, Yamasaki M. A case of ramsay hunt syndrome associated with local meningitis, multiple cranial neuropathy, and the second cervical nerve involvement. *Rinsho Shinkeigaku*. 1999;39:475-77.
- Doi H, Segawa F, Koyano S. A case of Vernet's syndrome due to varicella-zoster virus infection. *Rinsho Shikeigaku*. 2001;41:695-97.
- Patro S, Jena PK, Misra GC, Rath KC, Khatua P. Herpes zoster infection involving the maxillary branch of the right trigeminal nerve - a rare case report. *The Antiseptic*. 2013;110(1):36-8.
- Chitose SI, Umeno H, Hamakawa S, Nakashima T, Shoji H. Unilateral associated laryngeal paralysis due to varicella-zoster virus: virus antibody testing and videofluoroscopic findings. *J Laryngol Otol*. 2008;122(2):170-6.
- Nishizaki K, Onoda K, Akagi H, Yuen K, Ogawa T, Masuda Y. Laryngeal zoster with unilateral laryngeal paralysis. *ORL J Otorhinolaryngol Relat Spec*. 1997;59(4):235-7.
- Bossche VDP, Bossche VDK, Vanpoucke H. Laryngeal zoster with multiple cranial nerve palsies. *Eur Arch Otorhinolaryngol*. 2008;265(3):365-7.
- Watelet JB, Evrard AS, Lawson G, Bonte K, Remacle M, Cauwenberge VP, et al. Herpes zoster laryngitis: case report and serological profile. *Eur Arch Otorhinolaryngol*. 2007;264(5):505-7.
- Kim YH, Chang MY, Jung HH, Park YS, Lee SH, Lee JH, et al. Prognosis of ramsay hunt syndrome presenting as cranial polyneuropathy. *Laryngoscope*. 2010;120:2270-6.
- Patro S, Mishra GC, Toppo L, Katua P. Ramsay hunt syndrome with 8th cranial nerve palsy in a diabetic patient. *Orissa Physicians Journal*. 2010;6:98-102.
- Sahoo D, Patro S, Khora PK, Panda UN. A case of herpes zoster oticus with multiple cranial nerve palsy. *Int J Adv Med*. 2016;3:780-2.
- Korzec K, Sobol SM, Kubal W, Mester SJ, Winzelberg G, May M. Gadolinium-enhanced magnetic resonance imaging of the facial nerve in herpes zoster oticus and Bell's palsy: clinical implications. *Am J Otol*. 1991;12:163-8.
- Engstrom M, Abdsaleh S, Ahlstrom H, Johansson L, Stålberg E, Jonsson L. Serial gadolinium-enhanced magnetic resonance imaging and assessment of facial nerve function in Bell's palsy. *Otolaryngol Head Neck Surg*. 1997;117:559-66.
- Bossche VDP, Bossche VDK, Vanpoucke H. Laryngeal zoster with multiple cranial nerve palsies. *Eur Arch Otorhinolaryngol*. 2008;265(3):365-7.
- Ohashi T, Fusimoto M, Shimizu H, Atsumi T. A case of isolated vagus nerve palsy with herpes zoster. *Rhinso Shinkeigaku*. 1994;34:928-9.

Cite this article as: Patro S, Sahoo D, Dash AK, Mishra B. A rare case of herpes zoster maxillaris with cranial polyneuropathy. *Int J Adv Med* 2017;4:290-2.