

Herpes zoster involving the abducens and vagus nerves without typical skin rash

A case report and literature review

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Abstract

Rationale: Herpes zoster is characterized by unilateral vesicular eruption and it most often affects the trigeminal nerve. We would like to report a rare case of abducens and vagus nerves palsy caused by varicella zoster virus (VZV) without the typical vesicular rash.

Patient concerns: A 71-year-old woman presented with diplopia. Three days previously, she had experienced sore throat and hoarseness.

Diagnosis: At presentation, the prism cover test revealed esotropia of 10 prism diopters at primary gaze, and abduction was restricted in the right eye. No vesicular rash was observed on the patient's face, and magnetic resonance imaging of the brain showed no pathology. Flexible fiberoptic laryngoscopy revealed multiple ulcerations on the right side of the larynx—from the epiglottis to the arytenoid. After 1 day of hospitalization, a diffuse skin rash occurred on the patient's trunk, and polymerase chain reaction for VZV DNA was positive at the skin lesion. The patient was diagnosed as having herpes zoster associated with vagus and sixth nerve palsy.

Intervention and outcomes: She received famciclovir at a dose of 500 mg 3 times daily for 7 days and coadministered methylprednisolone. At the 4-month follow-up, her diplopia and eye movement had resolved completely.

Lessons: In patients with abducens nerve palsy without typical vesicular lesion, herpes zoster may not be detected early. In that case, systemic examination is very important for diagnosing herpes zoster.

Abbreviations: PCR = polymerase chain reaction, VZV = varicella zoster virus.

Keywords: abducens nerve, diplopia, vagus nerve, varicella zoster virus

1. Introduction

Herpes zoster is characterized by unilateral radicular pain and vesicular eruption caused by the varicella zoster virus (VZV).^[1] The most frequent sites of VZV latency are the trigeminal ganglia, although any autonomic ganglion or cranial nerve can be involved. VZV most often affects the sensory neurons, although the motor neurons can be affected in rare cases. In particular, there have been scattered reports of herpes zoster accompanied

by unilateral vocal cord paralysis, or by vocal cord paralysis and abducens nerve palsy.^[2]

Herpes zoster is difficult to diagnose clinically, especially when it manifests without the typical vesicular eruption. Herpes zoster involving other cranial nerves is often mistaken for more serious diseases, and thus resulting in extensive, unnecessary diagnostic workups that delay acyclovir treatment, which is only efficient when initiated within 72 hours from the onset of rash.^[3]

Herein, we describe the case of an adult patient with palsy of the abducens and vagus nerves. She presented with diplopia and hoarseness as isolated symptoms of central nervous system involvement during VZV infection without the typical vesicular rash.

Informed written consent was obtained from the patient for publication of this case report and accompanying images. No ethical approval was obtained because this study is retrospective case report and did not involve a prospective evaluation.

2. Case report

A 71-year-old woman presented with a chief complaint of diplopia, and sore throat and hoarseness lasting 3 days. She had no previous history of ocular disease or ocular surgery. The patient had well-controlled hyperthyroidism and no history of diabetes or hypertension. Upon examination, her best corrected visual acuity was 16 of 20 in both eyes. Her intraocular pressure, measured using a noncontact tonometer, was within normal limits. A slit-lamp examination was carried out and no ocular lesions were observed. A prism cover test revealed esotropia of 10 prism diopters at primary gaze, and that abduction was restricted

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Figure 1. Nine-gaze eye positions. Range of motion in the cardinal position demonstrates impairment of the lateral gaze (arrow) and nasal deviation at the primary gaze (arrow head) in the right eye.

in the right eye (Fig. 1). No vesicular rash was observed on her forehead or around her ears. Magnetic resonance imaging of the brain showed no pathology, and there was no evidence of acute microvascular ischemic disease.

The patient was admitted to the department of otolaryngology to evaluate her sore throat and hoarseness. Flexible fiberoptic laryngoscopy revealed multiple ulcerations on the right side of the larynx from the epiglottis to the arytenoid (Fig. 2). In addition, her right vocal cord was fixated in the paramedian position. She showed no tongue deviation. After 1 day of hospitalization, a diffuse skin rash occurred on the patient's trunk, face, and left knee, and viral exanthema was diagnosed in the dermatology department. A biopsy sample was taken from the skin lesion, and polymerase chain reaction (PCR) for VZV DNA was positive. The woman was diagnosed with herpes zoster involving unilateral vocal cord paralysis and abducens nerve palsy; she was prescribed famciclovir (3×500 mg/day for 7 days). To reduce inflammation, she was prescribed methylprednisolone (30 mg/day for 3 days, followed by tapering for 3 days). At the 4-month follow-up, her diplopia and eye movement had improved, and her vocal cord paralysis had completely resolved.

3. Discussion

VZV infection of the vagus nerve accompanied by abducens nerve palsy is extremely rare, and, to our knowledge, the present report is the first to record herpes zoster involving the vagus and abducens nerves without typical painful vesicles.^[4] In the present

case, the patient showed lateral gaze limitation of the right eye and fiberoptic laryngoscopy revealed a mucosal lesion that was confined to one half of the larynx, as we and the right vocal cord palsy, indicating that a branch of the vagus nerve was involved.

The mechanism of ocular motor involvement in VZV is still unclear. Several hypotheses have, however, been postulated.^[5–8] Specifically, Edgerton^[5] and Godtfredsen^[6] suggested that ocular motor involvement occurs via contiguous intracavernous radiculomeningitis, whereas Kreibitz^[7] posited that the extra-ocular palsies were due to perivasculitis–myositis rather than neural pathology. We speculated that these mechanisms all work simultaneously.

Herpes zoster can usually be diagnosed on the basis of characteristic pain and vesicular lesions. In patients with clinically suspected abducens nerve palsy without typical vesicular lesions, herpes zoster may, however, not be detected early. In such cases, systemic examination and history taking are crucial. Immunohistochemical examination, viral isolation tests, PCR for VZV DNA, and cell immunoassays are sometimes necessary to ensure differential diagnosis from other diseases.^[9,10] In the present case, we confirmed a diagnosis of herpes zoster using a PCR-based test of erosional exudate.

The appropriate treatment of VZV-related cranial nerve palsy is controversial. It is, however, important that antiviral agents such as acyclovir be initiated within 72 hours of the first symptoms.^[3] Such agents block the DNA synthesis in VZV at early-phase viral proliferation.^[11] Some authors have recommended using systemic corticosteroids to treat the possible

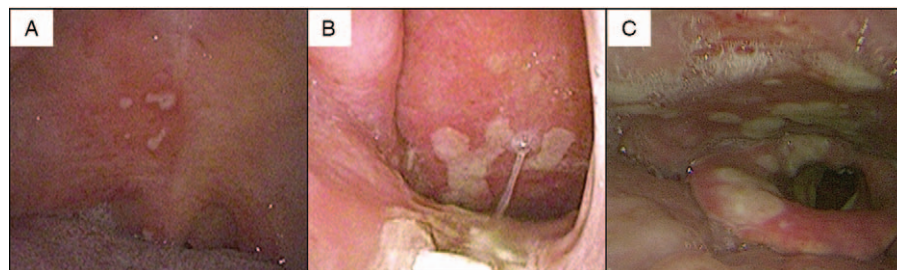


Figure 2. Flexible fiberoptic laryngoscopic images. Laryngoscopic image depicting mucosal exudates lateralized on the right side of the epiglottis (A), the posterior wall of the pharynx (B), and the arytenoid region (C).

vasculitic component of herpes zoster and/or prevent postherpetic neuralgia.^[12]

In the present case, 4 months after antiviral treatment was initiated, the patient's abducens and vagus nerve palsy had improved dramatically, and her diplopia had resolved. In general, the prognosis for spontaneous recovery of extraocular muscle function is favorable.^[13] The prognosis of vagal nerve palsy is, however, poor, especially when accompanied by glossopharyngeal nerve palsy, pharyngeal muscle weakness, and reduced sensation resulting in dysphagia and aspiration pneumonia.^[14] In the present case, the patient showed no tongue deviation, which means the vagus nerve palsy was not accompanied by glossopharyngeal nerve palsy. It may be for this reason that the patient's outcomes were so favorable.

4. Conclusions

In conclusion, in patients with clinically suspected abducens nerve palsy without typical vesicular lesion, herpes zoster may not be detected early. In such cases, VZV infection should be considered as the cause of abducens nerve palsy through systemic examination and history taking. In addition, early diagnosis allows prompt treatment and close follow-up, and thus suppressing viral proliferation in the early stages of the disease.

Author contributions

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