

A Case of Zoster Sine Herpete Misdiagnosed as Referred Otagia

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— ABSTRACT —

Patients with zoster sine herpete (ZSH) complain of deep and severe pain, which is limited to the dermatome without characteristic skin lesions. To our knowledge, there are few reports concerning ZSH wherein patients present with the chief complaint of otalgia. Here we report a case of ZSH that was originally misdiagnosed as referred otalgia at early stage. (*J Clinical Otolaryngol* 2015;26:77-80)

KEY WORDS : Zoster sine herpete · Otagia · Sudden hearing loss.

Introduction

Zoster sine herpete (ZSH) is one of the most difficult diseases to diagnose, since patients complain of deep and severe pain, which is limited to the dermatome.^{1,2)} Unlike herpes zoster oticus or Ramsay-Hunt syndrome, patients with ZSH do not exhibit characteristic skin or mucosal lesions in the external auditory canal, eardrum, or oral cavity. To our knowledge, there are few reports concerning ZSH wherein patients present with the chief complaint of otalgia. Here we report a case of ZSH that was originally misdiagnosed as referred otalgia.

Case Report

A healthy 27-year-old man with no medical history was referred to our outpatient clinic at the Eulji University hospital, complaining of continuous right-sided

otalgia. The otalgia had been present for 3 months, and sudden hearing loss on the right side had occurred 5 days before referral.

The otalgia was characterized by a continuous, deep, stabbing pain radiating from the right side of the head around the ear. The patient stated he had experienced flu-like symptoms 2-3 days before the onset of otalgia as well as a mild sore throat. Although he had been treated with pain medications after the initial diagnosis of pharyngitis-associated referred otalgia, the otalgia had not improved over the 3 months. During this time, the otalgia worsened, making daily activities and sleeping difficult, and ultimately culminating in hearing loss.

After a thorough physical examination, we found no signs of facial palsy or skin/mucosal lesions in the external auditory canal (EAC), eardrum, auricle, soft palate, or larynx. The patient did not exhibit signs of hypoesthesia or paresthesia in the face, cheek, or behind the ear. However, he did complain of worsening otalgia when the posterior parts of his EAC or retroauricular region were probed with fingers or forceps. Pure tone audiometry revealed total hearing loss on the right side.

Based on these observations, the patient was admitted to identify the causes of the otalgia and sudden hearing loss, since we suspected VZV infection, includ-

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ing ZSH. To treat the hearing loss, methylprednisolone (48 mg/day for 4 days, and tapered 8 mg every 2 days)

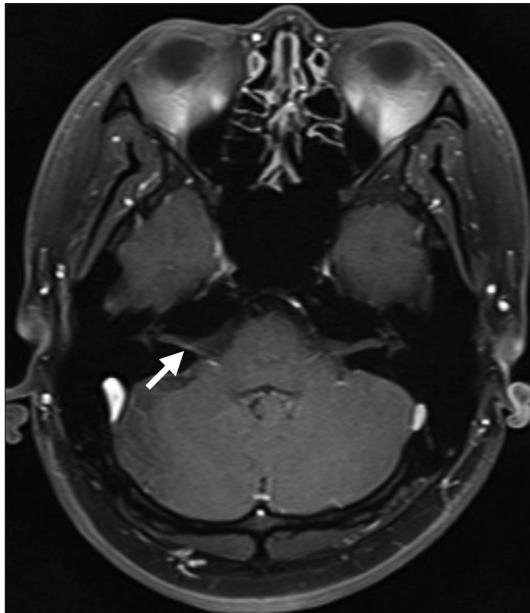


Fig. 1. Axial T1-weighted magnetic resonance imaging of the brain. The white arrow indicates a faint focal high signal on contrast-enhancement along the 7th and 8th intracanalicular nerve segment.

and valacyclovir (3,000 mg/day for 7 days) were prescribed. T1-weighted magnetic resonance imaging (MRI) of the brain revealed a faint focal high signal along the 7th and 8th right intracanalicular nerve segment, after contrast enhancement (Fig. 1). On the first day of admission, enzyme-linked immunosorbent assays (ELISA) and polymerase chain reaction (PCR) were used to detect the presence VZV antibodies (IgG and IgM) and DNA, respectively. ELISA results for VZV IgG were positive (1:13.65), although VZV IgM and DNA results were both negative.

Pain intensity was assessed daily using a visual analogue scale (VAS ; 0 : no pain, 10 : worse pain). The patient was treated with pregabalin, tramadol, amitriptyline, and capsaicin cream to control the severe otalgia, and the dosage and the timing of the medications are shown in Fig. 2. In addition, a stellate-ganglion block was inserted during the hospitalization period. On day 12 of hospitalization, pain intensity had decreased to 30% of the original severity, and the patient was discharged (Fig. 2). At this time, titers for VZV IgG and IgM were 1:10.06 and 1:0.40, respectively. After discharge, the patient received 3times of intra-

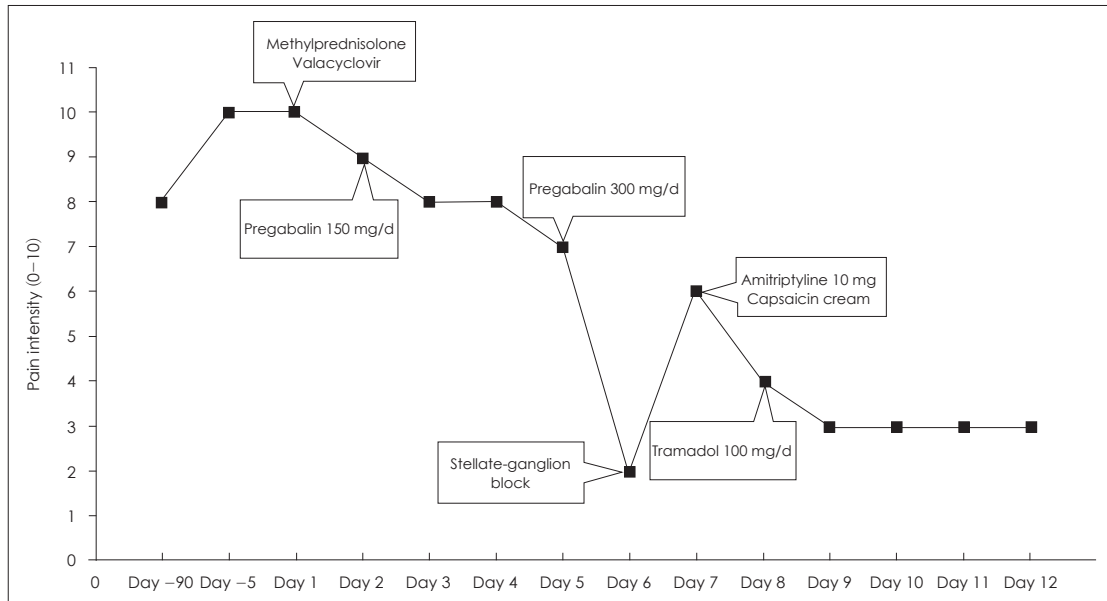


Fig. 2. Change in otalgia intensity over time. Treatments and dosages are indicated. Day 1 represents the first day of hospitalization.

tympenic dexamethasone injection over 2 weeks. After 1 month, the patient's hearing level showed no improvement, and VAS remained unchanged at a score of 3. However, for the titer of VZV IgG had decreased to 1:3.25, which was less than one-quarter of the initial titer of VZV IgG. Based on these results, the patient was finally diagnosed with ZSH.

Discussion

Otaglia can be classified as primary otaglia or referred otaglia. The common causes of primary otaglia include otitis media, mastoiditis, otitis externa, foreign body, and barotrauma. When primary otaglia is present, some abnormality of the outer or middle ear is frequently observed by physical examination.^{5,6)} On the other hand, the ear examination is usually normal in referred otaglia. Referred otaglia can be caused by temporomandibular joint disorder, dental problem, pharyngotonsillitis, head & neck cancers, temporal arteritis, and neuralgias involving cranial nerves V (trigeminal neuralgia), VII (geniculate neuralgia), and IX (glossopharyngeal neuralgia).⁷⁾ However, since our physical examination and dental consultation did not reveal any specific symptoms, these diseases were all excluded from the diagnosis. In addition, geniculate neuralgia was excluded, since neurovascular compression was not detected by MRI, as is the case in these patients.⁸⁾ In the physical examination, there are no skin rashes or vesicles, and if the character of pain is deep boring and superficial prickling, the possibility of ZSH must be considered.²⁾

To confirm VZV replication, VZV DNA (in saliva, serum, or cerebrospinal fluid), or a 4-fold increase in VZV titers (using paired sera) must be detected. In case of herpes zoster with vesicles, the sensitivity and specificity of PCR detection, using vesicular fluids reported at 95–99%.⁹⁾ However, the detection rate for ZSH remained unclear. We propose that the absence of detectable VZV DNA may be due to decreased viral replication over the 3-month course, or that blood samples may be not suitable for the detection of VZV DNA

in ZSH. Likewise, the decrease in VZV IgG titers may be attributed to the extended course, caused by the misdiagnosis of otaglia.

Unfortunately, we are unsure of the cause of this misdiagnosis. One possibility is that the initial physical examinations at local clinic may have revealed nonspecific physical findings except mild pharyngeal injection to obtain the original diagnosis. Another possibility is that small vesicular eruptions might have been overlooked at that time. Sadly, the course of disease in this patient was distinct from that of common herpes zoster oticus,⁹⁾ and we assume that an early administration of combined steroid-antiviral treatment might have altered the final outcomes.

With regard to treatment, this patient was treated with valacyclovir, although it seemed to be somewhat late, because he complained of severe otaglia as well as sudden hearing loss. These symptoms might be associated with ZSH, so we performed combined steroid-antiviral treatment considering both sudden deafness and acute zoster. In addition to VZV, other viruses including mumps, cytomegalovirus, influenza, Ebstein-barr virus, herpes simplex virus, and enterovirus can also cause sudden hearing loss.^{10,11)} However, antiviral treatment is not routinely used because the etiologies of sudden deafness remained unclear in most cases.¹²⁾ Similarly, the use of antiviral agent in postherpetic neuralgia is supposed to be ineffective for pain relief unlike acute zoster.¹³⁾ Instead, other medications including tricyclic antidepressants, gabapentin, pregabalin and topical lidocaine patch are more frequently prescribed.^{9,14)} Taken together, combined steroid-antiviral treatment is preferable in patients with sudden hearing loss accompanying severe otaglia.

Conclusion

In cases of severe otaglia with no abnormalities detected in the physical examination, the possibility of ZSH should be evaluated. Early detection and treatment may prevent the occurrence of adverse health outcomes.

REFERENCE

- 1) Kasahara M, Ichinohe T, Sano T, Fukuda K, Kaneko Y. *A case of zoster sine herpete of the trigeminal nerve. Bull Tokyo Dent Coll 2011;52(1):47-51.*
- 2) Lee HY, Kim MG, Park DC, Park MS, Byun JY, Yeo SG. *Zoster sine herpete causing facial palsy. Am J Otolaryngol 2012;33(5):565-71.*
- 3) Lee EJ, Yoon YJ. *A case of generalized varicelliform eruption after herpes zoster oticus. OtolNeurotol 2014;35(1):e57-9.*
- 4) Flint PW, Haughey BH, Lund VJ, Niparko JK, Richardson MA, Robbins KT, et al. *Otologic symptoms and syndromes. In: Cummings Otolaryngology Head & Neck Surgery, 5th ed. Philadelphia: Mosby;2010. p.2194-202.*
- 5) Park CW. *Geriatric Otolology: Overview. J Clinical Otolaryngol 2014;25(1):3-6.*
- 6) Yoo MH, Park HJ. *Diagnosis and treatment of Otalgia. Korean J Otorhinolaryngol-Head Neck Surg 2013;56(4):191-200.*
- 7) Ely JW, Hansen MR, Clark EC. *Diagnosis of ear pain. Am Fam Physician 2008;77(5):621-8.*
- 8) Tang IP, Freeman SR, Kontorinis G, Tang MY, Rutherford SA, King AT, et al. *Geniculate neuralgia: a systematic review. J Laryngol Otol 2014;128(5):394-9.*
- 9) Fashner J, Bell AL. *Herpes zoster and postherpetic neuralgia: prevention and management. Am Fam Physician 2011;83(12):1432-7.*
- 10) Schreiber BE, Agrup C, Haskard DO, Luxon LM. *Sudden sensorineural hearing loss. Lancet 2010;375(9721):1203-11.*
- 11) Kim YJ, Jung SH, Park BG, Mun MJ. *A Case of Mumps with Sudden Sensorineural Hearing Loss, Orchitis and Vestibular Neuronitis. J Clinical Otolaryngol 2014;25(2):255-60.*
- 12) Awad Z, Huins C, Pothier DD. *Antivirals for idiopathic sudden sensorineural hearing loss. Cochrane Database Syst Rev 2012;15:8:CD006987.*
- 13) Acosta EP, Balfour HH. *Acyclovir for Treatment of Postherpetic Neuralgia: Efficacy and Pharmacokinetics. Antimicrob agent chemother 2001;45(10):2771-4.*
- 14) Nalamachu S, Morley-Forster P. *Diagnosing and Managing Postherpetic Neuralgia. Drugs Aging 2012;29(11):863-9.*