A Letter

Oro-facial Herpes Zoster Affecting V2 and V3 Division of Trigeminal Nerve: An Unusual Case Report

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Abstract The reactivation of the Varicella Zoster virus (VZV) is called as Herpes Zoster (HZ) or shingles. Most commonly involved nerves are C3, T5, L1, L2 and first division of trigeminal nerve. This condition is characterized by multiple, painful, unilateral vesicles and ulcerations which show a typical single dermatome involvement. Herein, we are presenting a case of herpes zoster involving the V2 and V3 divisions of trigeminal nerve, showing unilateral vesicles over the left side of face, as well as an unusual presentation of involvement of the pre-auricular region and external ear. Intra-oral involvement of buccal mucosa, buccal vestibule, retromolar region, labial mucosa and the hard palate of the left side were evident; as also, loss of taste sensation in the anterior 1/3rd of tongue.

Keywords Herpes zoster; Trigeminal nerve; Vesicles; Oral ulcers

Introduction

Herpes Zoster (HZ) is an acute viral infection caused by reactivation of Varicella Zoster virus (VZV), following the primary varicella infection (chickenpox), usually in childhood (Owotude et al., 1999). It is characterised by inflammation of dorsal root ganglia or extra-medullary cranial nerve ganglia, associated with vesicular eruptions on the skin or mucous membrane in an area supplied by the affected nerve (Bandra et al., 2010). It is more commonly known as shingles, from the Latin cingulum, for ‘girdle’, because HZ involves a unilateral rash that can swathe around the waist or torso like a girdle (Roxas, 2006). Most commonly it results from a failure of the immune response to the latent virus, other factors such as radiation, trauma, medications, other infections, or stress can also trigger zoster (Mendieta et al., 2005). In our case, stress and fever were alleged factors for recurrence. HZ presents as a rash of 2-3 weeks’ duration in immunocompetent patients accompanied with moderate to severe pain. Oral manifestations of herpes zoster appear when the mandibular (V3), more commonly, and maxillary (V2), less commonly, divisions of the trigeminal nerve are affected. Generally, there is involvement of single nerve in majority of the cases. In our case, maxillary and mandibular divisions of trigeminal nerve were involved, along with skin and oral lesions, which was uncommon.

Case History

A 35 year old male patient reported to the Department with the chief complaint of blisters over the left side of face and ear as well as oral ulcerations since 3 days (Figure 1). History revealed that the patient had fever a week ago. He experienced burning sensation, followed by pain on the left side of the face as well as in the oral cavity. Gradually vesicles appeared 3 days back, which were initially few in number and later, increased to involve almost entire left half of the face and the...
external ear, up to the midline. The vesicles eventually ruptured with a watery discharge and were very painful. Patient had a history of chicken pox in childhood. Extra-oral examination revealed facial asymmetry. The skin on the involved side appeared shiny, swollen and tender, with multiple pinheaded, painful vesicles of varying sizes on the left middle and lower third of the face unilaterally, involving naso-lobial fold, infra-orbital area, upper lip, pre-auricular area and helix and anti-helix part of external ear, not crossing the midline (Figure 2). Intra-orally, multiple shallow irregular ulcerations, measuring approximately 5 to 10mm in size, with erythematous, irregular borders were noted on the buccal mucosa, buccal vestibule, retromolar area, hard palate and the labial mucosa, unilaterally, on the left side (Figure 3). Patient also experienced loss of taste sensation on the anterior 2/3rd of tongue. No dysphagia or odynophagia and facial palsy were reported. There were no other skin lesions accompanying the oro-facial lesions. The patient presented with vesicles on the external ear as well as loss of taste sensation on the anterior 2/3rd of tongue, however, James Ramsay Hunt syndrome was excluded as facial palsy was not evident. Hence, the diagnosis of herpes zoster involving maxillary and mandibular division was confirmed on the basis of classic clinical presentation.

Investigations: Hemoglobin: 13.5 gm/dl

- Random Blood sugar level: 84 mg/dl.
- Hepatitis B antigen test: Negative.
- ELISA test: Negative.
- Panoramic radiograph was taken to rule out any hard tissue finding due to dental or bony pathology, however, it did not reveal any significant finding (Figure 4).

Treatment: In our case, the patient was prescribed acyclovir, 800 mg five times a day and diclofenac sodium, 50 mg twice a day for 7 days along with viscous lidocaine mouth rinses on an as and when required basis. Patient was also prescribed prednisone, 10mg three times a day for 5 days. The patient responded well to the treatment showing healed lesions with scar and hypo pigmented areas (Figure 5). On the second appointment a week later, the patient was advised to continue with the topical application of acyclovir 1% ointment twice a day till the lesions healed completely (Figure 6).
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Figure 2 Multiple pinheaded, vesicles in relation to the left middle and lower third of the face not crossing the midline and involving the naso-labial fold, infra-orbital area, upper lip, pre-auricular area and helix and anti-helix parts of the external ear

Figure 5 Post-treatment profile photograph revealing complete resolution of the lesions intra-orally and healing of the lesions with scar formation extra-orally after 1 week

Outcome and Follow-Up: The patient was followed-up for 12 weeks after which the patient showed complete resolution of the lesions and no recurrences were reported.

Discussion
Varicella Zoster virus (VZV) is an omnipresent, DNA virus, which belongs to the subfamily human alpha herpes virus (Abendroth et al., 2010). The association between varicella and Herpes Zoster (HZ) was first made in the year 1892. The majority of HZ infections involve the thoracic and lumbar dermatomes; however, approximately 13% of the patients present with infections involving any of the three branches of the trigeminal nerve (Millar and Troulis, 1994). Varicella Zoster virus is responsible for two major clinical infections: primary (chicken pox- Varicella Zoster infection-VZI) and recurrent infection (shingles-Herpes Zoster infection-HZI). In 3-5 of every 1000
individuals, Varicella Zoster virus becomes reactivated causing lesions of localized herpes zoster with its incidence increasing with age and/or, immunosuppression in the affected individuals (Greenberg and Micheal, 2003). HZI of the skin (shingles) is more common in adults and starts with a prodrome of deep, aching and burning pain. There is usually little to no fever or lymphadenopathy. This is followed by the appearance of crops of vesicles in a dermatomal or zosteriform pattern within 2-4 days. The lesions usually begin to dry and scab 3-5 days after their first evident clinical appearance. Overall, the clinical course of the disease is usually between 7-10 days; however, complete healing may take more than 4 weeks (Sampathkumar et al., 2009). Involvement of the trigeminal nerve leads to the appearance of lesions on the upper eyelid, forehead and scalp with V1, midface and upper lip with V2 and lower lip with V3 involvement. With the involvement of V2, patients experience a prodrome of pain, burning and tenderness, usually on the palate, on one side. Also, it is a common observation that there is involvement of single nerve in most of the cases. The prevalence of involvement of both the nerves is uncommon and is seen only in 1.7% of the cases reported. In our case, the maxillary nerve (V2) and mandibular nerve (V3) were involved which was unusual. The diagnosis of Herpes Zoster is based on its archetypal clinical presentation. The diagnosis may be challenging in individuals during prodromal symptoms which persist for more than 2 to 3 weeks without any obvious clinical lesions, this phenomenon being known as “zoster sine herpete” (Stankus et al., 2000). In some patients, pain does not resolve when the rash heals but continues for weeks and sometimes, even for months or years later; the persistent pain being often termed as ‘post-herpetic neuralgia (PHN)’, one of the most commonly observed complications of the Herpes Zoster infection (HZI). PHN has been described as pain lasting 1-3 months after the complete resolution of the mucocutaneous lesions. PHN pain has been described as a pain that consists of three components: (i) a continuous, commonly deep pain; (ii) a brief, recurrent, shooting or electric shock-like “tic” pain; and (iii) a sharp, radiating, dysesthetic sensation evoked even by very light touching of the skin, termed as allodynia (Rowbotham and Fields, 1989; Basi-Kes and Demarin, 2007). Rare maxillofacial complications include developmental anomalies such as irregular short roots and missing teeth, periodontitis and calcified, degenerated pulps, indicative of non-vitality of the affected teeth (Bandral et al., 2010). Other potential complications of HZI include encephalitis, myelitis, peripheral nerve palsy and forms of contralateral hemiparesis. An uncommon complication of HZI involving geniculate ganglion is James Ramsay Hunt syndrome in which the patient develops Bell’s palsy, vesicles of the external ear and loss of taste sensation in anterior two-third of the tongue (Sampathkumar et al., 2009). In our case, the patient showed vesicles of the external ear and loss of taste sensation in anterior two-third of the tongue but facial palsy was not evident, thereby excluding the possibility of the abovesaid diagnosis.

References
Owotude et al., 1999