

Herpes Zoster Mandibularis: Dilemma of Diagnosis - A Case Report.

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ABSTRACT

Background: Herpes zoster infection involving the mandibular branch of the trigeminal nerve is rare. It can present with severe pre-eruptive pain along the nerve distribution. As presented by the patient in this study, this lesion may be mistaken for other painful conditions, leading to misdiagnosis and maltreatment. Post-herpetic neuralgia is the most common and important complication of herpes zoster infection. It is often refractory to treatment and can profoundly affect the patient's quality of life.

Objective: To report an unusual and rare case of herpes zoster mandibularis and the diagnostic dilemma experienced in the management.

Case Report: A 60-year-old female presented with severe intractable pain from the left side of the tongue and floor of the mouth, rashes of the lower lip and cheek, and ulcerations of the oral cavity. After a period of diagnostic puzzle due to confusing presentations, a diagnosis of herpes zoster infection involving the mandibular branch of the trigeminal nerve complicated with postherpetic neuralgia was finally made and the patient was treated accordingly. The peculiarity, management, and treatment outcome of the case are discussed.

Conclusion: Herpes zoster mandibularis, as seen in this case, can present with confusing clinical features and can lead to misdiagnosis and inappropriate treatment. This study aims to raise awareness of the clinical presentation of this rare entity among clinicians in medical and dental practices. It is also advisable to seek dental and orofacial evaluation for patients with a similar presentation from specialists in Oral Medicine and Maxillofacial Surgery

Keywords: Herpes zoster, mandibular nerve, postherpetic neuralgia, pain.

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Received: 16-Feb, 2023

Revision: 17 April, 2023

Accepted: 8 May, 2023

Citation: Akinbade AO, Oluwatuyi E, Busari OA. Herpes zoster mandibularis: dilemma of diagnosis - a case report. *Nig J Dent Res* 2023; 8(2):40-44. <https://dx.doi.org/10.4314/njdr.v8i2.1>

INTRODUCTION

Herpes zoster (HZ), commonly called *shingles* from the Latin *cingulum*, which means belt, presents as painful blistering skin eruption in a dermatomal distribution¹ and a distinctive syndrome caused by reactivation of varicella-zoster virus (VZV).

VZV usually causes chickenpox during childhood after which the virus remains latent in the sensory ganglia. It is the reactivation of the latent virus that causes Herpes zoster infection (HZI). It is characterized by prodromal symptoms such as fever, burning sensation and pain along the distribution of the involved dermatome. This pre-eruptive pain may precede the rashes by several days and may be mistaken for myocardial infarction, biliary or renal colic, pleurisy, dental pain, glaucoma, duodenal ulcer, or appendicitis, leading to misdiagnosis and maltreatment².

Postherpetic neuralgia (PHN) is pain after an acute episode of herpes zoster continuing beyond rash healing³. The pain has been described as a constant burning or stabbing sensation, while some individuals experience allodynia⁴. Symptoms can persist for months or even years, and the condition can profoundly affect patient's quality of life⁵.

Chen et al⁶ in a recent study done in Australia, reported a case of Herpes zoster mandibularis in an immune-compromised male which occurred after total parotidectomy. Faure et al⁷ in another study conducted in France also reported a case of mandibular osteonecrosis due to HZI of the mandibular branch of the trigeminal nerve in an elderly Caucasian male with uncontrolled and complicated diabetes Mellitus.

Owotade et al⁸ reported a case of HZI of the maxilla which presented with exfoliation of teeth and exposure of necrotic alveolar bone on the right side of the face. Fasina and associates reported a case series that highlighted the clinical presentation and management outcome of patients with herpes zoster ophthalmicus at a tertiary health facility in Southwest Nigeria⁹.

In a recent study, Olabode et al¹⁰ did an extensive literature review that examined the risk factors of herpes zoster and discussed different situations of its presentation with the illustration of cases involving the cervical nerve, thoracic nerve, and ophthalmic division of trigeminal nerve. Their study also discussed postherpetic neuralgia, other complications, treatment, and the future research trend of herpes zoster¹⁰. None of these previous Nigerian studies reported any case of herpes zoster

specifically involving the mandibular division of the trigeminal nerve. The aim of this study, therefore, is to report the unusual presentation of this rare case of herpes zoster mandibularis.

CASE REPORT

A 60-year-old woman presented in the Medical Outpatient clinic of our facility with severe intractable pain from the left side of the tongue and floor of the mouth of 5 months duration. She also had facial skin rashes and ulcerations of the mouth 2 weeks before the presentation. The pain was severe, and continuous and disturbed her sleep. She took many analgesics and applied herbal preparations and alcohol-based concoctions to relieve the pain which had markedly affected her quality of life as she appeared obviously depressed. She topically applied as much as 24 tablets (12,000mg) of paracetamol daily on the painful tongue for several weeks in search of relief. There was associated difficulty in mastication and weight loss since the onset of the pain but swallowing was not affected. She was tachypneic on presentation but other vital signs were within normal ranges.

Based on her presentation, features of dehydration, and the fact that the patient is a known diabetic, an assessment of immune suppression was made by the medical unit, while she was being investigated for oral candidiasis and squamous cell carcinoma of the tongue. She was subsequently referred to the Oral and Maxillofacial Surgery (OMS) clinic.

Further history taking at the OMS clinic revealed that the rashes were preceded by vesicles about two weeks earlier and on examination we found an apprehensive chronically ill-looking middle-aged woman, fully conscious but in severe painful distress. The retroviral screening was non-reactive, and HBsAg was negative, but the random and fasting blood glucose was elevated (11.7mmol/l and 8.6mmol/L respectively).

There was hypopigmentation and rashes on the left side of the lower jaw, as well as multiple ulcerations on the lips (Fig.1). The submandibular lymph nodes were not tender, temporomandibular joints were palpable and not tender but mouth opening was limited (2.5cm maximum inter-incisal distance). There were perioral ulcerations and rashes involving the upper and lower lips. The oral hygiene was very poor and there were non-bleeding adherent darkish white plaque-like coatings on the dorsal and ventral surfaces of the tongue, floor of the mouth, and part of the palate (Fig.2). The tongue and the floor of the mouth on the left side were also exquisitely tender to

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touch. There was no muscle or nerve dysfunction on the face. Based on the history of vesicles and other features above, a diagnosis of herpes zoster infection involving the mandibular division of the trigeminal nerve (Herpes zoster Mandibularis) complicated by postherpetic neuralgia was made.

Following this diagnosis, the patient was immediately administered recommended dose of

tabs Biopentin (Gabapentin 300mg and methylcobalamin 500mg) for the first week, tabs Celecoxib 200mg 12hrly and tabs 100mg vitamin C 8hrly for one and two weeks respectively. All previous medications except metformin and glibenclamide were discontinued.

Figure 1



Clinical photograph of the patient on presentation.

Figure 2



Clinical photograph of patient 1 week after the commencement of treatment.

When the patient came for review a week after the commencement of treatment, she reported drastic and significant relief of pain. Her appearance showed

marked improvement, she had no fresh complaint and the tenderness of the tongue and floor of the mouth had markedly subsided (fig. 2). She could now

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feed and sleep well. The rashes and ulcers were healing, the oral hygiene has improved and the whitish coating on the tongue and floor of the mouth were clearing (fig.2). Biopentin was continued for another two (2) weeks and it was ensured that the dosage was titrated at the beginning and tapered at the end as appropriate. The patient sustained clinical improvement as the pain, perioral rashes and

ulcerations impressively resolved and her quality of life drastically recovered (fig. 3).

Even though the patient did not show up for the scheduled recall visit one month after treatment she was regularly followed up through telephone calls and she has been free from the symptoms of the disease for over 2 years after the treatment.

Figure 3



Clinical photograph of the patient 2 weeks after commencement of treatment.

DISCUSSION

Herpes zoster affects the trigeminal nerve in approximately 13% of cases, and in most of these cases, the ophthalmic and maxillary divisions are involved. The involvement of the mandibular division of the trigeminal nerve (V3) is the least common¹¹. There is general paucity of data on Herpes zoster affecting the mandibular nerve. Authors are not aware of any clinical case report on herpes zoster mandibularis from Africa in the English literature^{10,11}. Reactivation of the VZV, in this case, maybe due to advancing age and impaired immunity due to Diabetes Mellitus. The first peculiarity of this case was the unusually long duration of the pre-eruptive pain the patient had for over four months. This duration is longer than several days or less than or equal to 3 days reported by Gnann² and Forbed et al¹² respectively in the literature. The whitish coating on the tongue which also extended to the right side was a confounding feature that contributed to the diagnostic dilemma as it mimicked oral candidiasis. This could have been caused by the alcohol-based concoction and paracetamol tablets that the patient applied on the tongue for many months before the

presentation. The pain from the tongue which has been associated with oral cancer¹³ coupled with the history of weight loss and ulceration could make the diagnosis of squamous cell carcinoma of the tongue a consideration. However, the weight loss here could have been caused by difficulty with mastication and uncontrolled diabetes mellitus the patient presented with.

Another critical feature of this case is that the severe pre-eruptive pain continued even weeks after the eruption of the vesicles. This is suggestive of early complication with postherpetic neuralgia in this patient. An antiviral drug like acyclovir or valaciclovir which has been effective for treating herpes zoster^{6,9} was not administered to the patient because the acute phase of the herpes zoster infection was considered to have elapsed on presentation. The rapid response of the patient to biopentin without antiviral medication may have confirmed our impression. Other studies in agreement with this current report, have shown that tricyclic antidepressants and gabapentin either as a single agent or in combination are effective in treating post-

herpetic neuralgia by reducing the severity and duration of the pain^{9,10}

The diagnostic dilemma of this case was not only a result of the peculiarities stated above but also the rarity of the involvement of the mandibular branch of the trigeminal nerve in herpes zoster infection as confirmed by the lack of publication of case reports on herpes zoster mandibularis from Nigeria. The negative impact of this condition on the quality of life and the susceptibility of the patient to substance/drug abuse is enough reason to advocate a routine vaccination scheme against VZV in Africa like in other developed nations¹⁴.

CONCLUSION

Herpes zoster mandibularis is a rare condition that can present with bizarre clinical features that may be misdiagnosed to be oral candidiasis, squamous cell carcinoma and some other pain disorders. Reactivation of the VZV in this case may be due to advancing age and impaired immunity due to diabetes mellitus. The duration of pre-eruptive pain in this case was unusually long and the transition to post-herpetic neuralgia was comparatively shorter. Biopentin therapy administered for 3 weeks was effective in this patient.

The purpose of this study is to raise awareness of the clinical presentation of this rare entity among clinicians in medical and dental practices. It is also advisable to seek dental and orofacial evaluation for patients with a similar presentation from specialists in Oral Medicine and Maxillofacial Surgery.

Source of support

Nil

Conflict of interest

None Declared

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