

A rare case report and appraisal of the literature on spontaneous tooth exfoliation associated with trigeminal herpes zoster

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Abstract

Background Reports of post herpetic maxillofacial complications have been very rarely documented in the literature that includes periapical lesions, calcified and devitalized pulps, resorption of roots, osteonecrosis, and spontaneous exfoliation of teeth. The atypical feature of the case of concern to the dental surgeon is the rare complication of spontaneous tooth exfoliation following herpes zoster.

Case report This case reports a male patient of age 47 years who reported to the Department of Periodontology with the chief complaint of mobility in the left upper central incisor. Patient history revealed herpes zoster infection that began 11 days earlier along with underlying diabetes mellitus condition. We hereby report a known diabetic patient with history of herpes zoster infection who presented with rare complication of spontaneous tooth exfoliation involving the maxillary division of the trigeminal nerve.

Discussion Limited number of cases has been reported in the literature regarding spontaneous teeth exfoliation secondary to

herpes zoster. The exact pathogenesis regarding the spontaneous exfoliation of teeth in herpes zoster patient is still controversial. Thus, an oral health care provider should be aware of this rare complication while managing a case of tooth mobility with the previous history of herpes zoster of trigeminal nerve.

Keywords Herpes zoster · Tooth exfoliation · Trigeminal nerve · Post herpetic

Introduction

The varicella-zoster virus is a ubiquitous human alpha herpes virus, which causes two clinical entities varicella (chicken pox) and herpes zoster (shingles). Varicella (or chickenpox) is a benign illness of childhood as a result of the primary infection of varicella-zoster virus. It is a highly contagious infection and is characterized by an exanthematous vesicular rash. When the varicella infection occurs, virus in the skin travels up the sensory nerves to become latent in the sensory ganglia [1, 2]. Once the varicella-zoster virus in its latent state is reactivated in the dorsal root ganglia, it results the inception of herpes zoster (HZ) infection that results in a localized cutaneous eruption [3]. Herpes virus (herpes, creeping) describes the nature of the pathologic lesion [4]. Little is known about the mechanisms that account for preservation of latency and subsequent reactivation of the virus. The spread of virus occurs through nerves to the skin, and a vesicular cutaneous eruption develops in the area innervated by the affected sensory nerve after the reactivation and active viral replication in the sensory ganglia. Among the cranial nerves affected by herpes zoster infection, trigeminal nerve is the most commonly affected (18.5 to 22 % of total cases), followed by glossopharyngeal nerve and hypoglossal nerve. It is conceivable that intraorally, symptoms appear when the second

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(maxillary) or third (mandibular) trigeminal divisions are affected [2, 5, 6].

Reports of post herpetic maxillofacial complications have been very rarely documented in the literature that includes periapical lesions, calcified and devitalized pulps, resorption of roots, osteonecrosis, and spontaneous exfoliation of teeth. Spontaneous exfoliation of teeth in the area innervated by the affected nerve has been reported rarely in the literature. It is considered to be an early [7, 8] episode by some researchers, while others consider it as delayed [5] complication of herpes zoster infection. We hereby report a case of spontaneous tooth exfoliation in a patient suffering from herpes zoster infection with underlying diabetic systemic condition.

Case description

A 47-year-old male patient reported to the Department of Periodontology, Himachal Dental College, Sundernagar, with the chief complaint of mobility in the left upper central incisor. Patient also complained of numbness and tingling sensation in the upper left quadrant. Extraoral examination revealed residual crusty lesions of herpes infection over the left side of the face restricted to the distribution of the maxillary division of trigeminal nerve with no associated lymphadenopathy was noticed. There were also areas of healing with scar formation and hyperpigmentation (Fig. 1 and Fig. 2). On intraoral examination, slight redness of the alveolar mucosa was seen around the left upper central incisor along with grade III mobility with no alterations in the remaining teeth of the upper left quadrant (Fig. 3). Patient history revealed herpes zoster infection that began 11 days earlier with typical itching and paraesthetic sensation. After 3 days, vesicles appeared on an erythematous zone affecting the skin over the left side of face. The patient was being treated with oral (800 mg/daily for 10 days) acyclovir for the herpes zoster infection. Patient past medical history exhibits his underlying diabetes mellitus condition.

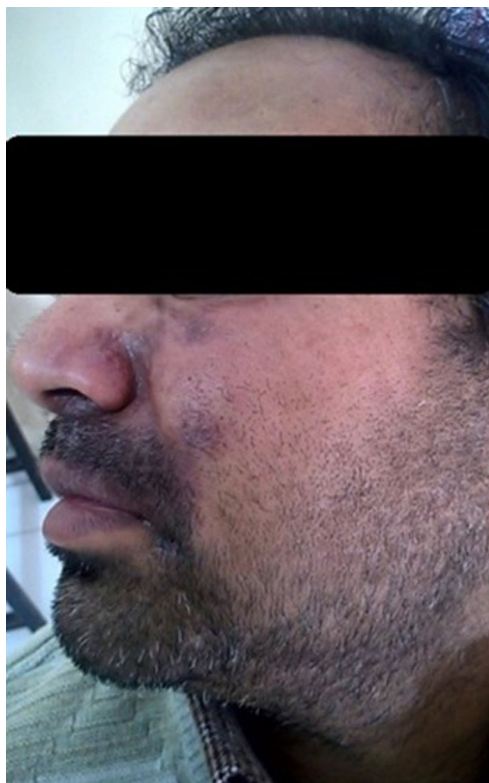


Fig 2 Frontal view showing scar formation and hyperpigmentation

He was on medication for the same from the past few years. He was asked to bring all the medical records, but the patient reported next day with exfoliated upper left incisor tooth. The intraoral examination revealed normal alveolar socket with no purulent exudates. The radiological examination revealed empty tooth socket on the left side of the maxilla with no sequestrum (Fig. 4).

Routine hematologic test investigations revealed marked lymphopenia with reactive lymphocytes present, suggestive of viral infection. Other laboratory investigations were within the normal limits (HIV, hepatitis B, and routine urine analysis). The values of fasting blood sugar were slightly raised



Fig 1 Scar formation and hyperpigmentation as seen on the left side of the face



Fig 3 Intraoral view showing normal alveolar socket without purulent exudates

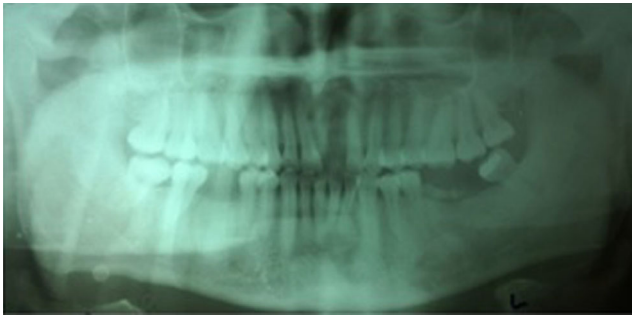


Fig 4 Orthopantomogram (OPG) showing empty tooth socket on the left side of maxilla with no sequestrum

(140 mg/dl). The patient was prescribed on antibiotic therapy (amoxicillin 500 mg, three times daily for 10 days) and was instructed to rinse with 0.12 % chlorhexidine digluconate.

Discussion

Herpes zoster affects all ages, but has been seen to be rare in children and adolescents [2]. It has no gender predilection but there is a slight male predominance [2, 5, 6], and the onset commonly occurs among individuals in the sixth through the eighth decades of life (five to 10 cases per 1000 persons) [5]. The incidence of HZ infection increases with age and in general population has been reported to be 5.4 % [9] while it has found to be up to 10 times higher in individuals suffering with HIV infection [10]. Moreover, studies suggest that prevalence of herpes zoster infection increases among immunocompromised patients such as patients infected with HIV infection, hematologic malignant disease, immune-mediated disorder, and organ transplant patients, and the risk of herpes zoster infection for such immune-suppressed patients also increases according to the age [6, 11, 12]. Other risk factors take account of external damage of the affected dermatomes, psychological stress, and race [12]. Among the affected dermatomes, it has been found that most commonly affected dermatomes are the thoracic, cervical, and trigeminal dermatome [11].

Intraorally, the lesions of herpes zoster appear when the mandibular or maxillary divisions of the trigeminal nerve are affected. The lesions of herpes zoster infection can appear on the face, in the mouth, in the eye, or on the tongue when the branches of the trigeminal nerve are involved [2]. The herpes zoster infections usually undergo three stages that includes the following: (1) Prodromal stage presenting as the sensations described as burning, tingling, itching, boring, prickly or knife like occurring in the skin over the affected nerve distribution which is usually preceded by the rash of the active stage by few hours or several days. (2) Active stage (or the acute stage) characterized by the emergence of the rash which progresses from erythematous papules and oedema to vesicles in 12 to 24 h and finally progresses to pustules within 1 to 7 days. The

intraoral vesicles, scattered and surrounded by an erythematous zone, soon become ulcerated and covered by a white pseudo membrane. The vesicles dry and crust over after 1 to 2 weeks, but it may take several weeks for the skin to return to normal. (3) Chronic stage also termed as post herpetic neuralgia is the most significant complication of herpes zoster infection [2, 3, 8, 13, 14].

Besides herpes zoster infection, other predisposing conditions have included HIV/AIDS, malignancy, chemotherapy, radiation therapy, and diabetes mellitus [15]. Published reports have demonstrated increased incidence of herpes zoster in diabetic patients [16, 17]. In diabetic patients, occlusion of the vessels leading to tissue ischemia is present with associated predisposition to infection. Impairment of the neutrophil function particularly chemotaxis can be another contributing factor [15, 18]. It has been hypothesized that the increased risk for herpes zoster among patients with diabetes mellitus may also be related to decrease varicella zoster virus (VZV)-specific cell-mediated immunity. Yet more relevant studies are required to find the exact mechanism by which diabetes mellitus affects VZV-specific cell-mediated immunity [17]. The case reported in this paper had the medical history of diabetes which could be one of the predisposing factors to spontaneous tooth exfoliation.

The maxillofacial complications of herpes zoster infection including the spontaneous exfoliation of the teeth and the osteonecrosis have been described as rare phenomena [2, 6, 19]. These cases have rarely been reported in the literature, and a summary of the available data on patients with trigeminal herpes zoster infection associated with the exfoliation of teeth has been given in Table 1.

The first case concerning the relation of osseous alterations with the herpes zoster infection was reported in the literature by Rose in 1908 [2]. In 1922, it was Dechaume [20] et al. that accredited Gonnet as the pioneer to have reported on this rare complication. The enigma still remains regarding the time interval amid the outbreak of herpes zoster infection and the spontaneous exfoliation of teeth in the area innervated by the affected nerve. Published reports have demonstrated that spontaneous tooth exfoliation and bone necrosis occur as early as the first 2 weeks of initial infection while other authors consider this to be a delayed complication that will occur between the third and twelfth weeks after the onset [5, 7, 8, 15, 21, 22]. In the present case, tooth exfoliation occurred within 2 weeks of initial herpes zoster infection.

The underlying rationale for the spontaneous exfoliation of teeth can be attributed to the alveolar bone necrosis and/or to necrosis of the periodontal ligament [2, 23]. The uncertainty still exists regarding the process by which the HZ infection leads to the alveolar bone necrosis. Some researchers believe that it could be due to local vasculitis caused by direct extension of the neural inflammatory process to adjacent blood vessels and infarction of trigeminal vessels that accompany

Table 1 Review of the available data on patients with trigeminal herpes zoster infection associated with the exfoliation of teeth

S.no	Author/Year	Age/Gender	Systemic disease/Treatment	Site of necrosis	No. of teeth lost
1.	Dechaume et al ,1955 [20]	48/M	Chronic hepatitis, anemia, leukopenia	L. mandible	5
2.	Delaire and Billet ,1959 [26]	71/F 79/F	NP NP	R.mandible R.mandible	1 6
3.	Hall et al ,1974 [27]	62/F	Reticulum cell sarcoma (nasopharynx) , radiation therapy	R.maxilla	7
4.	Chemitz 1976 [28]	76/M 85/M	No major illness NP	L.maxilla R.mandible	3 5
5.	Vickery and Midda 1976 [29]	41/F	Disseminated Hodgkin's, stage IV-B ,chemotherapy	L.mandible	7
6.	Cooper 1977 [25]	76/M 85/M	No major illness NP	L.maxilla R.mandible	3 5
7.	Delbrouck-Poot and Reginster 1979 [30]	44/M	Hodgkin's disease	R.maxilla	5
8.	Schwartz & Kvorning 1981 [8]	66/F	No major illness	R.mandible	4
9.	Wright et al ,1983 [24]	56/F	Histiocytic lymphoma, stage IV-B , chemoradiation therapy	L.maxilla	7
10.	Manj et al,1986 [31]	60/F	Polymyalgia rheumatica	R.mandible	2
11.	Mostofi et al,1987 [32]	56/NP	Leukemia ,chemotherapy	R.mandible	3
12.	Toshitaka et al ,1990 [23]	72/NP	NP	R.mandible	2
13.	Peñarrocha et al, 1992 [33]	50/M	NP	R.maxilla	7
14.	Mintz and Anavi 1992 [21]	50/M	NP	R.maxilla	7
15.	Owotade et al,1999 [34]	45/M	No Major Illness	R.maxilla	2
16.	Mendieta et al 2005 [2]	63/F	No Major Illness	R.mandible	2
17.	Siwamogstam et al , 2006 [22]	30/F 31/M 29/M 31/F	HIV HIV HIV HIV	L.maxilla R.mandible R.mandible L.mandible	2 1 1 2
18.	Shabnum meer et al, 2006 [15]	70/M	Diabetes, CMV infection	L.mandible	3
19.	Jain et al,2010 [5]	65/M	Tuberculosis	R.mandible	7
20.	Kim NK et al,2012 [6]	78/M 77/M	No major illness No major illness	R.mandible R.mandible	2 3
21.	Lambade P et al,2012 [35]	52/M	NP	L.maxilla	2
22.	Mahajan VK, 2013 [36]	80/M 55/F 45/M	No major illness No major illness No major illness	R.maxilla R.mandible R.mandible	Incisors till molars Total loss of teeth Total loss of teeth
23.	Cloarec N et al ,2014 [37]	50/M	HIV	R.mandible	2
24.	Patil S and Al Zarea BK ,2015 [38]	58/M	HIV	R.maxilla	2
25.	Okumara K et al ,2015 [19]	72/M	Meningoencephalitis	L.mandible	1
26.	Present case	47/M	Diabetes	L.maxilla	1

Modified and updated from Wright et al [24]

NP: not provided, HIV: human immunodeficiency virus, CMV: cytomegalovirus

the trigeminal vessels supplying the jaws [24] or generalized infection of the trigeminal nerves supplying the periosteum and periodontium of the involved dermatome area [7]. Perhaps an ischemic problem with these attributes could result in more than one tooth affected [2], while others are of the opinion that pre-existing pulpal or periodontal inflammatory conditions or surgical procedures performed in the site of zoster infection [24, 25] or systemic viral infection of the

odontoblasts causing degenerative tissue changes that could contribute to more destructive alveolar bone necrosis [5].

The management of herpes zoster infection includes treatment with antiviral agents that have been shown to reduce the duration of the rash and severity of pain associated. Yet it has been reported that the benefits have only been seen in patients who were prescribed antiviral agents within 72 h after the onset of the rash [10]. The most significant complication of

herpes zoster infection is post herpetic neuralgia which is generally a self-limited condition. The most debilitating complication of herpes zoster is osteonecrosis which is a rare phenomenon. Timely commencement of antiviral therapy and aggressive analgesic treatment can prevent this post herpetic complication of osteonecrosis. In cases of secondary infection with osteonecrosis, sequestrectomy along with the removal of inflammatory tissue and regular patient follow-up must be done [2, 5, 6]. When osteonecrosis of the jaw is accompanied with tooth exfoliation as rare complications, then active use of painkillers, regulation of topical factors, and proper extraction of dead bone and affected teeth can lead to better treatment outcomes [6].

In the present case scenario, the patient had no periapical lesion or periodontal involvement as depicted on radio graphical examination yet the authors are of the opinion that patients underlying diabetic condition may have contributed to the spontaneous tooth exfoliation along with herpes zoster infection. Thus, an oral health care provider should be aware of this rare complication while managing a case of tooth mobility with the previous history of herpes zoster of trigeminal nerve.

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