Case Report

Severe mandibular osteonecrosis and spontaneous teeth exfoliation after an outbreak of herpes zoster infection - a case report from surgical approach to prosthetic rehabilitation

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Abstract

Herpes zoster, also known as shingle, is an infection caused by Varicella-zoster virus (VZV), and it results from reactivation of endogenous latent VZV infection within the sensory ganglia. There are several complications of herpes zoster, but only limited reports of osteonecrosis with teeth exfoliation have been addressed. In this case report, we documented a prosthetic reconstruction utilizing the tilited implant in severe mandibular osteonecrosis and spontaneous teeth exfoliation after an outbreak of herpes zoster infection. Tilting implant with angled abutment was selected combining with straight implants, which provided an alternative and simple solution for post-shingle outbreak patient associated with an extensive osteonecrosis and teeth exfoliation to effectively reconstruct the oral masticatory function.

Introduction

Herpes zoster, also known as shingle, is an infection caused by Varicella-zoster virus (VZV), and it results from reactivation of endogenous latent VZV infection within the sensory ganglia. The virus spread along with the nerves to the associated dermatome causing vesicular eruptions. There are several complications of herpes zoster, but only limited reports of osteonecrosis with teeth exfoliation have been addressed. In this case report, we documented a prosthetic reconstruction utilizing the tilited implant in severe mandibular osteonecrosis and spontaneous teeth exfoliation after an outbreak of herpes zoster infection. Briefly, the surgical intervention was successfully associated with a good prognosis despite the accidental mandibular fracture, which was also fixed at that time. During the regular follow up, no further osteomyelitis or infection was noticed. The patient was in good compliance and well maintained regular follow-up after the treatment. Thus, he was transferred to our Prosthodontics Department for further prosthetic reconstruction. During comprehensive diagnostic workup for oral rehabilitation, we noticed the difficulty of conventional approaches in this case due to the anatomical limitation in the mandible. Tilting implant with angled abutment was selected combining with straight implants, which provided an alternative and simple solution for post-shingle outbreak patient associated with an extensive osteonecrosis and teeth exfoliation to effectively reconstruct the oral masticatory function.

Case report

A 58-year-old HIV-negative male came to our family dentistry department in December 2012 for evaluation of the facial swelling which had persisted for a while. According to his statement, he visited a local dental clinic due to a toothache and asking for routine teeth scaling. After scaling, the dentist prescribed with ibuprofen for pain control, which accidentally caused his allergic reaction. Then, he presented at our Dermatology Department in November 2012, with a chief complaint of a painful, reddish swelling sensation associated with a scab of the upper and lower lips as well as the right lower cheek. The dermatologist considered an allergic reaction. But teeth 44 and 45 spontaneously exfoliated after few days follow-up. He, therefore, was diagnosed with herpes zoster combined erythema mutiforme. After medication with Famvir and weekly monitoring, the patient was transferred to our family dentistry for further evaluation within a week.

Extraoral examination revealed a reddish swelling and scab on the lips and the right lower cheek. Intraoral examination showed a significant mandibular bony exposure around the socket and the distal extension around exfoliated teeth. Tooth 48 was mobile with Miller's grade II mobility. The remaining teeth were vital, with fair oral hygiene, though a mild to moderate of periodontal disease was noticed. Panoramic radiographic revealed a prominent massive radiolucency shadow around the right mandibular body. Moreover, we noticed a dramatically distal infiltration of osteonecrosis within two days as compared with the difference of panoramic films. Furthermore, another exfoliation of tooth 48 was seen during the next visit.

The patient was immediately referred to the Oral and Maxillofacial Surgery Department for further management. After two days follow-up, herpes zoster infection related osteonecrosis of the right area of the mandible was considered, and sequestrectomy was arranged. The patient was admitted on December 27th, 2012. The operative sequestrectomy of right lower mandible under general anesthesia was performed on the next day. Laboratory data was normal from the pre-operative investigation. The whole procedure of debridement of necrotic bone was successful, and postoperative recovery was uneventful. The patient was discharged after 3-day post-operative care. The postoperative wound healing was uneventful. However, the patient presented to the Oral and Maxillofacial Surgery Department on January 9th, 2013 because of accidentally biting a chicken bone. Physical examination showed tenderness and extraoral swelling on right mandibular angle and submandibular area without intraoral swelling. Furthermore, the panoramic x-ray revealed an oblique fracture line at the right mandibular angle. He was admitted immediately. An open reduction and internal fixation with intermaxillary fixation were performed under general anesthesia. The whole operative procedures were successful, and he was discharged two days later. During his regular follow-up and no further evidence of infection or exfoliation of the tooth was noticed.

Prosthetic Rehabilitation

The patient presented and asked for further prosthodontics treatment after regular follow-up appointments and the osteonecrosis and exfoliation teeth were stable. In his first appointment, preliminary impressions with irreversible hydrocolloid were made. The study casts were mounted on the articulator with the aid of a face-bow. Intraoral examination revealed only teeth 33, 34, 37, and 38 were remaining. The crossbite of teeth 33 and 34 was noticed. Limited remaining teeth was possibly the factor causing the crossbite. However, the occlusion was stable with the remaining teeth. The mylohyoid muscle was detached during the surgery. Thus, a significant elevation of mylohyoid muscle from the lingual side was observed during the movement of the tongue. At the buccal side, the extremely shallow vestibule and the lack of keratinized gingiva were noticed. The maximum mouth opening was within normal limit. The panoramic radiograph revealed the amount of mandibular bone in posterior right side was limited. Conventional partial removable prosthesis could not achieve the patient's expectation due to unfavorable bony, teeth, and soft tissue support. Intraorally, in his right side of the mandible, there was limited prosthetic space without enough vestibule or keratinized gingival tissue. On the other side of the jaw, the remaining teeth were questionable to serve as abutment teeth due to mild mobility and poor crown-root ratio.

Diagnostic teeth arrangement on the base plate for the lower arch with wax up for tooth 37 was performed for surgery reference. Because of the crossbite with remaining lower anterior teeth, the

space of lower anterior area was wider than that of in normal population. We, therefore, placed five lower anterior teeth to achieve better esthetics and designed an edge-to-edge relationship to reach a stable mutual-protective occlusion. Four simulated dental implants were located at 46, 43, 41 and 32 positions. After checking the wax denture in the patient's mouth and reviewing the panoramic radiograph and CT scan, a surgical stent was fabricated, and implantation surgery was arranged.

Four dental implants (Nobel Biocare, Sweden) were placed on mandible based on surgical guide under general anesthesia in June 2014. We chose to place three straight implant and one distally titled implant due to limited bone height. The tilted implant was placed around the area of projected tooth #46, connected with 30-degree multi-unit abutment. All other three implants were connected with straight multi-unit abutments, covered with healing caps. Simultaneously, the bone plate and screw previously placed for fixation were removed. The surgical procedures were smooth and successful, and the wound healing was uneventful. Due to the fact that the primary stability of less than 30Ncm was achieved, the conventional implant loading protocol[1] was decided to adapt.

The definitive impression at the abutment-level was performed with an open-tray, splinted technique six months later. The wax rim retained with two screws were used for bite registration. Wax denture was successfully applied to check occlusion, fitness, and esthetics. Acrylic teeth with high impact pink acrylic were fabricated with one screw. After confirming the occlusion, the rest of the screws were picked-up with titanium prosthesis abutments. The prosthesis was fixed with hand-torque, and the screw holes were sealed. The patient was received oral hygiene instruction reinforcement with dental flossing and interdental brush to be able to clean around the implant-supported prosthesis. Weekly follow-up was arranged in the first month. The patient occasionally complained cheek biting while chewing in the first and second week follow up. After slightly adjustment of the mandibular right canine tip, that situation was significantly improved. Proper oral hygiene was observed in every follow-up appointment. The patient tolerated well of the prosthesis throughout the whole observation period. The 4-year follow-up showed the stable and pleasant outcome of the implant-supported prosthesis.

Discussion

Osteonecrosis of the jaws is an infrequent complication of herpes zoster infection. Only 41 cases of severe osteonecrosis of the jaws have been reported. In this case report, the first case discovered in Taiwan have been completely diagnosed, treated, and monitored. In the general population, the incidence of herpes zoster infection has been reported to be 5.4 %[2]. The incidence has been reported 0.3%-1% among the people older than 80 years[3]. In Taiwan, from 2000-2008 annual incidence of herpes zoster was increasing gradually, from 4.45 per thousand cases to 6.89[4]. Moreover, the incidence rate increases for those experience varicella infection in childhood, especially in those experience varicella before two years old[5]. The infection of herpes zoster may affect cranial nerve, but the trigeminal nerve is the most frequently affected[6]. The unilateral infection is the unique feature. In our case, the shingle, the area of osteonecrosis and exfoliation of teeth are at the same unilateral right mandibular distribution.

The mechanism of the osteonecrosis followed by teeth exfoliation from herpes zoster infection remains to be disclosed. Several hypotheses have been proposed. Two possible factors of the pathogenesis mechanisms were concluded in Mendieta's review[7], including (1) ischemic necrosis affecting localized dermatome area and (2) local inflammation at involved area causing tissue necrosis. Besides, (3) preexisting plural or periodontal inflammatory condition or surgery performed in the infected zoster site might aggravate bone necrosis[8]. However, in a recent study, the majority of patients were edentulous, indicating local dental problems might not be the primary factor[9]. Osteonecrosis could affect the periodontal tissue such as alveolar bone as well as the periodontal ligament. Consequently, this pathological scenario results in the shedding of the tooth[10]. Spontaneous exfoliation of teeth usually occurs in the first two weeks of the infection[11]. Nevertheless, this severe complication should be aware by dentist and physicians in the future.

In our case, the patient denied having any systemic disease or any predisposing factor such as HIV or immunosuppression disease. However, after the breakout, severe osteonecrosis followed by spontaneous three teeth exfoliated approximately occurred within ten days. Antiviral medication therapy and keeping the intraoral wound clean by irrigation daily were successful in controlling

the further damage. Surgical debridement was necessary to limit the spread of the bone necrosis. While most of the cases remained missing of prosthetic reconstruction after wound healing, our patient was cooperative, maintained regular follow-up after the treatment. Thus, he was transferred to our Prosthodontics Department for further prosthetic reconstruction.

To our best knowledge, the complication of mandibular fracture after herpes zoster infection was only reported once before our current report[12]. In the previous case, mandibular fracture due to pathologic factors was suspected. However, in this case, the fracture happened due to accidentally biting into chicken bone. The sequestrectomy results in the weakened mandible and causing fracture at mandibular angle.

After sequestrectomy, the amount of posterior right side of the mandible bone was limited. A conventional partial removable denture could not fulfill patient's expectation due to the unfavorable bony, teeth, and soft tissue support. Intraorally, in his right side of the mandible, there was limited prosthetic space without vestibule or keratinized gingival tissue. On the other side of the jaw, the remaining teeth were questionable as abutment teeth due to mild mobility and poor crown-root ratio. Thus, the implant-support prosthesis was the best-fit in this circumstance. To achieve an adequate implant height, we utilized one tilted implant and three straight with the advantage of cross-arch stability to restore the oral and masticatory function.

In Guido's systemic review[13], for a partial fixed dental prosthesis, a 5-year and 10-year prosthetic with a respectively 98.9%, 97.8% survival rate were estimated on 2-4 implants prosthesis. Though it was tough to compare and evaluated the varieties of replaced-units to implant ratios, the ratio of 2/3 was recommended to achieve an adequate result in that review. In our case, it was impossible to replace the total of nine teeth with six implants due to the anatomical limitation. Implants distribution was also improved by utilizing the tilted implant.

The typical All-on-4 concept provides a reliable surgical and prosthesis protocol with a high success rate to overcome anatomical limitations[14,15]. Typical All-on-4 was used for full arch rehabilitation. However, we used the tilted implant in this case for the partial fixed prosthesis. A predictable good result was expected with the improvement of the implant distribution. According to Fabbro's review, an excellent outcome can be expected regarding short and medium prognosis[16]. Moreover, using tilted implantation allowed the longer implant to be placed, and avoided using short implants or bone grafts. Moreover, it enhanced the load distribution and made it possible to use the all-acrylic prosthesis. Nevertheless, long-term prognosis should be evaluated carefully in the future.

When encountering atrophic bone deficiencies, the straight implant may have difficulty in achieving adequate length for osseointegration. In Zampelis's analysis, distal tilting of implants splinted by fixed prosthesis did not increase the stress to the bone[17]. Furthermore, using the tilted implant could result in a favorable soft tissue seal[18]. In our case, there was an incident soft tissue dehiscence with the straight implant abutments at first two months after the implant surgery. The patient was unable to keep good oral hygiene at the beginning of the healing stage. That might be the possible factor influencing the healing process. oral hygiene reinforcement and wound irrigation with chlorhexidine during the follow-up appointments, soft tissue began to heal with implant abutment. By keeping an adequate level of oral hygiene, using the tilted implant for partial fixed denture provides a predictable technique for prosthetic rehabilitation[19]. However, long-term study is minimal and more evidence need to support this technique in the future.

Using metal ceramics is considered as the gold standard for the fabrication of the implant-supported prosthesis. Recently, the CAD/CAM technology provided an innovative way of using inexpensive material, and predictable manufacturing outcome can be achieved. In this case report, the acrylic prosthesis was selected due to the limitation of the CAD/CAM technology in our facility back in 2014. The advantage of using acrylic is that it is easier to repair, and the material is relatively lower cost. The complication of using acrylic is the wear of occlusal surface and catastrophic fracture[20]. The other consideration of using multiple abutment is the fatigue of the abutment screw. However, no complication was noted since the prosthesis was delivered.

In summary, in this case report, we have illustrated the severe oral complications of herpes zoster infection including, osteonecrosis, exfoliation teeth, and mandibular fracture. The progress of the complications was extremely rapid and severe. This shingle outbreak results in severe complications should be aware both in

medical and dental professionals to prevent further damage for future treatment strategy and monitor. Moreover, the post-operative care of sequestrectomy should be concentrated on avoiding potential mandibular fracture and recovering and reconstructing the masticatory function. Taken together, in this case report, we provided a successful treatment strategy and complete prosthetic rehabilitation for shingle related osteonecrosis followed by tooth.

Figure 1. The extraoral view of the herpes zoster infected site.

Figure 2. The intraoral view of the herpes zoster infected site.

Figure 3. The close-up view of the herpes zoster infected site.

Figure 4. The panoramic radiograph of the implant placement. Noted that fixation plate was inserted in the right side of the mandible.

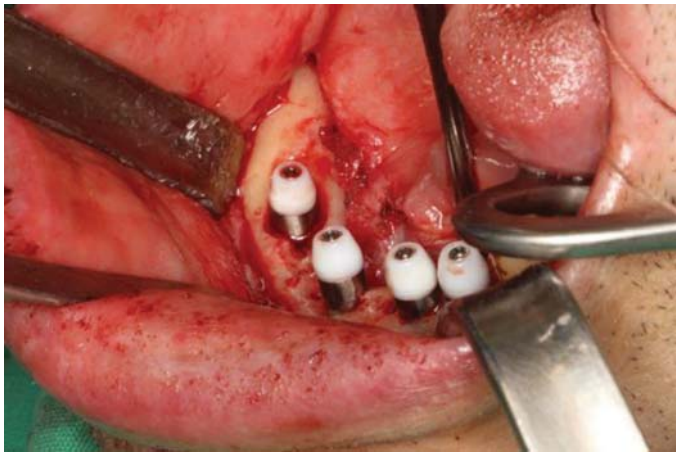

Figure 5. The intraoral view of the implant placement.

Figure 6. The two week follow-up after the implant placement.

Figure 7. Frontal view of the acrylic prostheses.

Figure 8. Front view of the prosthesis at the 4-year follow-up.

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