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Case Report

Management of palatal perforation in systemic lupus erythematosus patient

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ABSTRACT

Background: *Systemic Lupus Erythematosus (SLE) is an autoimmune disease which damages tissues and causes chronic inflammation with an idiopathic etiology. It has been suggested that oral lesions in patients with SLE can be grouped clinically as erythema, discoid lesions and oral ulcerations. The latter have been said to indicate the onset of a severe systemic disease flare and that oral ulcers represent cases of mucosal vasculitis. Palatal lesions generally present in the form of ulcers or, in more severe forms, as perforation. Acquired palatal perforations can be caused by several etiologies including: developmental disorders, malignancy and infections.* ***Purpose:*** *To report the management of palatal perforation in an SLE patient.* ***Case:*** *A 14-year-old female patient attended the Dr. Ramelan Naval Hospital, with both a perforated palate that often caused her to choke when eating or drinking and maxillary anterior tooth mobility.* ***Case Management:*** *The treatment for the patient in this case consisted of debridement and DHE, pharmacological therapy including aloclair gel and minosep mouthwash to maintain oral hygiene and prevent re-infection. At the end of the first consultation, the patient was prescribed an obturator in order to avoid oro-anthral infection. During the second consultation, the patient's orthodontic bracket was removed to facilitate scaling and splinting of the anterior maxillary teeth carried out to prevent their movement. During the third consultation, a swab was taken by an oral surgeon who also administered antifungal therapy. During the fourth and final consultation, the patient was examined a prosthodontic specialist due to an obturator which was causing discomfort.* ***Conclusion:*** *The management of palatal perforation lesions in an SLE patient requires a multidisiplinary approach.*

Keywords: *systemic lupus erythematosus; palatal perforation; oral manifestation; candida; obturator*

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INTRODUCTION

Systemic lupus erythematosus (SLE) constitutes a chronic systemic autoimmune disease[1,2] whose precise etiology remains unexplained. However, there are certain pre-dispositional factors underlying it, including: genetics, infection, hormones, antibodies, the immune system, sunlight, diet, stress and physical fatigue.[2] SLE can present itself in a wide variety of ways. It may affect one or more organs, being characterized by widespread inflammation of the blood vessels and connective tissues, and tends to be episodic with intermittent periods of remission. According to the American College of Rheumatology (ACR), a key symptom of SLE is the presence of a malar rash affecting both cheeks known as a facial butterfly rash[3] which is usually accompanied by joint and muscle pain, fatigue, hair loss (alopecia), anemia, oral ulcers, high fever, decreased appetite and chest pain or chest tightness among other symptoms. While individual patients present contrasting symptoms, the presence of four of these support a diagnosis of SLE[3,4] which can attack parts of the body such as the skin, oral cavity, joints, blood and even the internal organs,

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for example the kidneys and lungs to name but a few. The type of illness and organs affected differ from one patient to another with the result that it is referred to as an individual disease.[4,5]

Approximately 75% of people with SLE present various symptoms in the oral cavity including an oral ulcer located in the buccal mucosa, palate, periodontitis with floating teeth and a dry mouth accompanied by a burning sensation, especially when eating spicy or hot foods.[1–3,5] According to Sete, *et al*.[1] several studies exist confirming the association between chronic periodontitis and SLE conditions, characterized by damaged periodontal tissue resulting in floating teeth and ulcers on the oral mucosa, and most of them on the hard palate.

A diagnosis of SLE can be established through anamnesis, characteristic clinical manifestations and supporting investigation, such as complete blood tests and an antinuclear antibody (ANA) test. From this examination, the existence of the autoimmune process can be determined.[4] The main treatment for SLE is the administering of corticosteroids as an immunosuppressant because when the condition occurs the pre-dispositional factors result from abnormalities in the immune system. Further therapy may consist of anti-malarial drugs or other symptomatic treatment. In immunocompromised states, long-term treatment with corticosteroids and antibiotics may cause the microorganisms to develop into pathogenes.[6] Corticosteroid therapy should be conducted under the supervision of competent medical personnel due to the side effects of long-term treatment with corticosteroids including: osteoporosis, elevated blood sugar levels, hypertension (resulting in a full moon face) in addition to the risk of infection, both viral and bacterial, and fungal infections.[7]

The most common type of fungus found in the oral cavity is candida two strains of which, *Candida albicans* and *non-Candida albicans* (NCAC), cause fungal infection referred to as oral candidiasis. 84.8% of cases of oral candidiasis, including: *Candida crusei*, *Candida tropicalis*, *Candida parapsilosis*, *Candida kefyr*, and *Candida famata,* are caused by *Candida Albicans* and NCAC.[8] In recent years, the prognosis for individuals suffering from SLE has improved considerably when treated with appropriate therapy and the greater ease of access to both information and the health services equipped to treat the condition.[2] The purpose of this paper was to report the management of palatal perforation in SLE patients which can provide insight for dentists working in an integrated multidisciplinary manner in other fields in order to offer optimal care in SLE cases requiring holistic treatment.

CASE

A 14-year-old female referred by the Prosthodontics Department attended the Oral Medicine Department at Dr. Ramelan Naval Hospital in Surabaya together with her father. The purpose of this prosthodontic referral was to ascertain whether a patient diagnosed with SLE could undergo a safe palate perforation involving the creation of an obturator, since the prosthodontist himself had been referred by an oral surgeon. The patient's chief complaint was that of frequent choking while eating and drinking due to palate perforation, although she experienced little pain. Her father stated that between January and March 2017 the patient had already been hospitalized at RSUD Bojonegoro with dengue fever on four occasions. Her blood test results confirmed that she had experienced a decrease in both platelets and hemoglobin.

On March 17 2017, the doctor treating the patient referred her to Dr. Ramelan Naval Hospital in Surabaya where she was admitted as an in-patient. The provisional diagnosis was one of suspected hypovolemic shock and febrile neutropenia with pancytopenia. The patient had undergone a complete blood examination and ANA test at the Clinical Pathology Laboratory of Dr. Ramelan Naval Hospital, Surabaya, the results of which supported a diagnosis of SLE.

When the patient was hospitalized at Dr. Ramelan Naval Hospital, rashes suddenly developed on both cheeks which, while not itchy or tender themselves, were accompanied by painful cracked lips. In early April, following the patient's discharge from hospital, a red ulcer appeared on her palate which, on April 25, turned white. Despite an increase in the

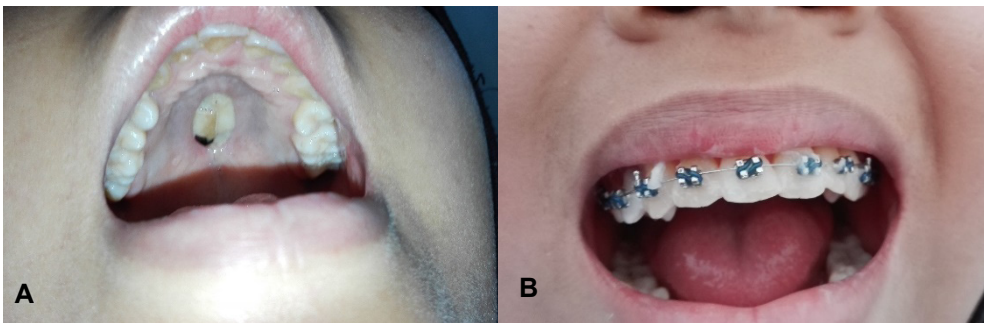

Figure 1. A) Palate middle: Ulcer, with painful *bone exposure*. B) Patient with a fixed orthodontic brace.

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ulcer's size, the fact that it was pain-free led to its being regarded as merely an ordinary in nature. By the end of June 2017, the ulcer had a needlepoint hole which gradually grew to its current size (Figure 1A, 1B).

On extra-oral examination, a blackish ulcer was found on the skin of the hand. The patient, who had been fitted with a removable orthodontic appliance, was also found to be suffering from lymphadenopathy. Intra-oral examination of the palate region revealed an ulcer and necrotic exposure of the palate bone which caused considerable pain. Moreover, the maxillary front teeth appeared to be mobile. Although her general condition appeared to be relatively good, the patient appeared somewhat depressed.

The patient had been placed on a range of medication by various doctors. The internist had prescribed methylprednisolone, chloroquine, folic acid, sandimmune, and Cavit-D3; the oral surgeon mefenamic acid and cefadroxil; and the psychiatrist based at RSUD Bojonegoro a concoction of mood-enhancing medicines.

CASE MANAGEMENT

Visit I (July 6th, 2017): According to the medical records, on the first visit, notes existed on the results of the panoramic photo, blood test and ANA test previously conducted. An ANA test examination confirmed the patient to be ANA IFM positive as determined by previous diagnoses of SLE cases manifested in the oral cavity. Management of the condition consisted of a combination of debridement and Dental Health Education (DHE), while pharmacologic therapy combined the application of aloclair gel (aloe vera extract) to reduce pain and minosep mouthwash to maintain oral hygiene and prevent secondary infection.

In order to address prosthodontists' uncertainty regarding whether the use of obturators as a symptomatic and rehabilitative therapy was permitted, the authors, as oral medicine specialists, sanctioned their manufacture. Instructions governing the use of obturators stated that they should not be used continuously but, rather, only during eating in order to avoid the possibility of an allergic reaction should the patient suffer from an autoimmune disease. Another piece of advice stated that the obturator must be removed and cleaned at night to avoid fungal infections potentially exacerbating the patient's existing condition.

The treatment plan to be implemented in SLE cases included several essential steps. First, implementation of an integrated approach involving cooperation with other specialists such as periodontists, orthodontists, prosthodontists and oral surgeons. Second, removal of the orthodontic bracket. Third, completion of scaling and splinting of the teeth in question. Fourth, taking an impression for the manufacture of an obturator. Fifth, swabbing the mouth in order to produce a tissue culture as a means of identifying the microorganisms present. Sixth, referral to an anatomical pathology specialist for assessment

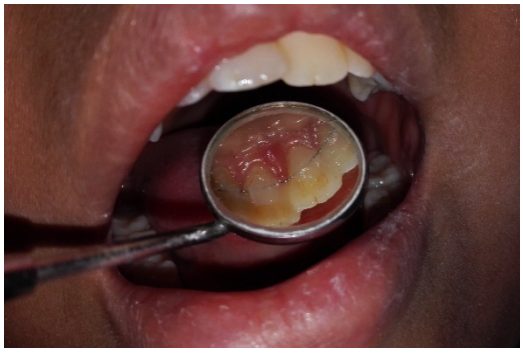

Figure 2. The results of splinting using palatal wire.

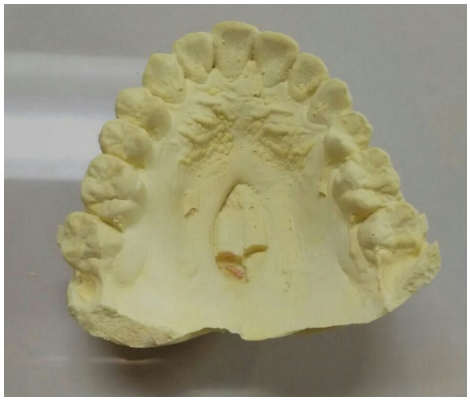

Figure 3. A patient's dental impression.

of the condition of the surrounding tissue. Lastly, regular attendance of the control by the patient.

Visit II (July 7th 2017): The processes of anamnesis and clinical examination conducted during the second consultation were identical to those of the patient's first visit. In accordance with the original plan, the management on the second day included: removal of the orthodontic bracket, scaling and splinting work in the resorbing region (Figure 2) involving teeth 11, 12, 13, 21, 22, and 23 (loose teeth with 2-3 degrees of mobility), and taking an impression for the manufacture of an obturator (Figure 3).

Visit III (July 10th. 2017): On the third visit (second control), the patient's pain had been reduced due to her having obeyed fully all the instructions issued during the previous consultation. On this occasion, management consisted of performing a swab examination in order to identify the type of fungus present which was conducted by an oral surgeon experienced in the investigation of fungal infections. In the case reported here, the presence of a deep fungal infection was suspected because of the existence of perforation. Consequently, the patient was administered an antifungal therapy involving fluconazole 200 mg which is regarded as the gold standard for such treatment. In the final step, an obturator was inserted.

Visit IV (July 18th 2017): On the fourth visit, the anamnesis conducted confirmed that the patient felt more comfortable with the obturator, despite the continued

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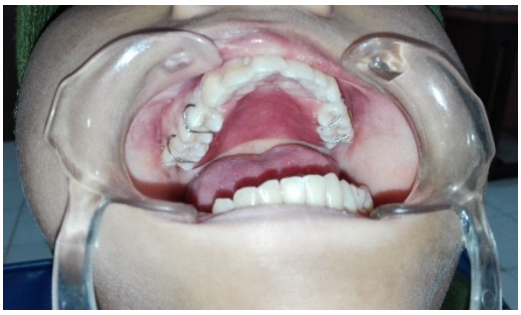

Figure 4. Application of an obturator by a prosthodontist.

leakage she experienced when eating or drinking. Her swab result confirmed the presence of both *Candida crusei* and *Candida tropicalis*. The subsequent patient management involved a prosthodontic check-up to correct the functional use of the obturator (Figure 4). The final step consisted of referring the patient to the Anatomy Pathology Department in order to ascertain the recent condition of the lesion on her palate.

DISCUSSION

Infection constitutes the entry of microorganisms into the tissues or body fluids, accompanied by both local and systemic clinical symptoms, which can occur on the skin, in soft tissues, in ulcers and in surgical wounds. Infectious diseases caused by a variety of viruses, bacteria and fungi are a major cause of morbidity and mortality.[9]

In the past, fungal infections have been continually underestimated. Therefore, in the present day, they should attract serious attention not only because of a failure to diagnose them correctly, but also due to their potential to lead to higher mortality. The incidence of fungal infection is classified as an endemic condition, for example: histoplasmosis, blastomycosis, coccidioidomycosis, paracoccidioidomycosis and opportunistic fungal infections.[10] Fungal infections that are primarily opportunistic might demonstrate severe progression, particularly in patients with immunocompromised conditions including: diabetes mellitus, malignancy and SLE among others.[4]

In the oral cavity, this species is found commensally in 40% of the intra oral healthy population and dominates 90% of isolates from patients with *Candida albicans.*[4] Although the most frequently found species is *Candida albicans*, recently the incidence of NCAC infection has continued to increase.[8] Over 30% of SLE patients often present problems with the salivary gland where an unstimulated saliva flow rate results in decreased salivary production and, in turn, severe Sjögren's syndrome, oral candidiasis and xerostomia.[2]

Oral ulcers, painless lesions often found in the buccal mucosa and palate, have been identified as the major diagnostic manifestation of SLE by the American Rheumatism Association Committee on Diagnostic Therapeutic Criteria. Such ulcerative lesions are usually small (less than 1 cm), although SLE-related oral lesions observed in the palate tend to be longer and larger, resembling red plaques surrounded by white areas.[2] The patient's major complaint in this case was that of an oral complication arising from palate perforation which caused her discomfort when eating or drinking because she often started to choke. From analysis of the swab taken, the presence of *Candida crusei* and *Candida tropicalis* was confirmed. Similarly, an anatomical pathology examination revealed the role of fungi and bacteria.

The possibility of etiopathogenesis from growth of the ulcer to perforation of the palate was investigated through several processes. As previously explained, the intact mucosa constitutes one of the body's innate immunity system against fungal infections. When an ulcer is present in a patient's mucosa, the mucosa is no longer intact - a condition which facilitates invasion by microorganisms. Long-term use of corticosteroid therapy results in the emergence of *Candida tropicalis* and *Candida crusei* which both belong to the *Candida albicans* family. These generally cause diseases in tropical climates, especially in patients with leukopenia, neutropenia and immunocompromised conditions, as in the case of SLE, and facilitate the development of a fungemia pathogen.

Candida tropicalis is one NCAC species demonstrating the highest level of virulence due to its attachment ability, the strongest among epithelial cells, which can secrete protein at medium levels.[8] The fungus entering the body induces a response from the host's immune system. IgM and IgG are produced and circulated by the bloodstream in response to fungal infections. In fact, these two kinds of candida are commensal in nature, but they can develop into pathogens. In the presence of fungal exposure, this phenomenon can depend on the type and degree of immune response from the host. Cellular immune responses are a major mediator of the body's resistance to fungal infections. Neutrophils and phagocytes play an important role in eliminating fungi.[10]

Sete, *et al.*[1] state that, with regard to SLE, there are several possible causes such as: disruption of T cell function, genetic defects in the immune response control system, abnormal macrophage functioning, B cell damage, ineffective host response to an infectious agent or a combination of several of these elements.

Candida tropicalis and *Candida crusei* are primarily commensal in healthy individuals, but can cause oral candidiasis accompanied by painful symptoms in immunocompromised individuals. In the case of SLE, because of a compromised immune system highlighted by Sete, *et al.*[1], severe damage may result in defects in the bone palate which can develop into necrosis potentially culminating in perforation of the bone palate. Kumar, *et al.*[11] added that, in SLE, the lesion initially

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appears as an irregular whitish area that then extends towards the peripheral, after which its center reddens and develops into an ulcer with a hyperkeratotic edge.

In this case, according to the anamnesis obtained from the patient's closest blood relative (her father), she initially had a reddish ulcer which then turned white and grew in size until, finally, a small hole appeared in the palatal mucosa. This gradually expanding hole occurs due to deficient tissue healing and the failure of the patient's immune system which enables the candida to develop into a pathogen. This statement is supported by the immunocompromised condition of the patient diagnosed with SLE, an autoimmune disease, which renders the tissue more easily infected.

The treatment for the patient in this case consisted of provision of the debridement and DHE, pharmacological therapy using aloclair gel and minosep mouthwash to maintain oral hygiene and prevent a second infection after the first visit. At the end of that visit, the patient used an obturator in to order to avoid an oro-anthral infection. During the second visit, the patient's orthodontic bracket was removed and scaling and splinting of the anterior maxillary teeth performed to prevent movement of the anterior maxillary teeth. The third visit witnessed a swab being undertaken and antifungal therapy administered by the oral surgeon. Although nystatin and amphotericin b are the drugs most commonly used locally, fluconazole oral suspension is proving to be very effective in treating oral candidiasis. Fluconazole was found to be the drug of choice for the systemic treatment of oral candidiasis.[12] During the final visit, the last patient consulted the prosthodontic specialist due to an obturator which was causing discomfort.

In addition to the reduced host immune response, SLE was suspected based on several symptoms, including xerostomia, characterized by a dry mouth which results in reduced saliva production. Since saliva contains several antifungal elements such as lysozyme, histatin, lactoferrin, and calprotectin, its reduced production causes a decrease in cellular and humoral immunity.[13]

SLE disease is an autoimmune disease that affects many women of reproductive age with clinical symptoms affecting many organs, one being the oral cavity. The chief complaints arising from palate perforation include: impaired intake processing of food, difficulty in talking or communicating and the production of a nasal sound during speech. Moreover, the ongoing infection caused by the disease leads to halitosis. All of the above effects can undermine the self-confidence of children and, indeed, the teenage patient who was the subject of this case study was very concerned about her appearance.[2]

SLE disease affecting the oral cavity can negatively impact the quality of life of the patient. Therefore, the handling of oral manifestations of this disease should be undertaken carefully to ensure that patients receive proper care supportive of the success of the treatment. In conclusion, commensal candida may develop into pathogens in immunocompromised hosts such as SLE.

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