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A RARE CLINICAL SYPHILIS ORAL MANIFESTATION DUE TO SEXUALLY TRANSMITTED DISEASE AMONG HOMOSEXUAL: CASE REPORT

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ABSTRACT

Background: Syphilis is a sexually transmitted disease caused by *Treponema pallidum*, often found in Human Immunodeficiency Virus (HIV) patients and homosexuals. The oral manifestations are chancres, mucous patches, focal epithelial hyperplasia (FEH) and papilloma/condyloma. FEH is a rare oral mucosa disorder that occurs in immunocompromised patient. **Objective:** To present a rare oral manifestation in syphilis patient due to sexually transmitted infections. **Case:** A 25-year-old homosexual male came to Oral Medicine Department complaining of sores on the palate for 3 weeks and 2 weeks later a painless lump appeared. He was an HIV patient and had been taking ARVs since 6 months ago. Pain when opening the mouth, itching on his face and genital area. Intraoral revealed a soft, pedunculated, non-bleeding, mobile mass at the palate with a diameter about 2 cm, had the same color as the surrounding tissue. There was a reddish band-like area on the maxillary anterior gingival margin, suspected as Linear Gingival Erythema (LGE). **Case management:** 0.2% Chlorhexidine gluconate mouthwash was administered to prevent secondary infection and he was referred to Dermatovenereology and Periodontology Department. HBsAg Immunochromatography (negative), VDRL titer (reactive), TPHA titer (reactive) therefore diagnosis of syphilis was made and he was treated with a single dose Benzathine penicillin G 2.4 million units intramuscular. The oral lesions healed after 1 month therapy. **Conclusion:** FEH is a rare oral mucosa disorder in syphilis patients. Knowledge of the signs and symptoms of the disease is needed to make a diagnosis and a multidisciplinary approach to achieve successful treatment.

Keywords: Oral manifestation, Sexual transmission, Syphilis, Treponemal infection

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INTRODUCTION

Sexually transmitted diseases (STDs) are worldwide public health problem and becomes a cause of death if do not get proper treatment. The exact incidence and prevalence of STDs in many countries are not known. According to the World Health Organization (WHO), 250 million new cases of STDs, including gonorrhea and syphilis, are diagnosed each year.^{1,2}

Syphilis is a sexually transmitted disease caused by *Treponema pallidum* and is frequently found in individuals with the Human Immunodeficiency Virus (HIV).^{3,4} Schaudinn and Hoffmann identified *Treponema pallidum*, an anaerobic bacteria with a diameter of 0.1 – 0.18 micrometers and a length of 6 – 20 micrometers, as the causative agent of syphilis in 1905.^{3,5,6}

Globally every year estimated 6 million new cases of syphilis in people aged 15 to 49 years; more than 300,000 fetal and neonatal deaths are associated with syphilis; and 215.000 infants die prematurely.⁵ According to WHO, in 2016, it was estimated that

globally the prevalence of syphilis was 0.51% in men.⁷ A study in 2019 in 30 EU/EEA countries reported 270,000 cases of syphilis with male-to-female ratio of 8.6:1. This disease is often found in the population aged 25 – 34 years and over 45 years.⁶ Data on the incidence of syphilis in Western Europe, the United States and China show that the male population that has sex with men (MSM) is higher than that of heterosexual men.^{4,5} Based on estimates done by the Ministry of Health in 2012, there were are around 9 million people in Indonesia who were at high risk of contacting or transmitting HIV, and of these, there were approximately 1.15 million men who have sex with men (MSM). In 2007, 2009, 2011, and 2013, the Ministry of Health periodically conducted the Survei Terpadu Biologi dan Perilaku (STBP). The STBP results showed that HIV prevalence was increasing among MSM, with a 9% prevalence of syphilis cases among MSM.⁸ Syphilis became a condition that remained remarkable in the MSM population and in groups that tended to have multiple (heterosexual) sex partners.⁵

Generally, syphilis is transmitted sexually through direct contact with an infected person; although this is considered safe, the use of condoms or oral sex can also be a way of transmitting syphilis.⁵ Syphilis can also be transmitted congenitally, when spirochetes pass through an infected woman's placenta, enter the fetus's body, and infect it. Syphilis can also be transmitted through blood donation.^{5,6} After an average incubation period of 3 weeks (range 10 – 90 days), the lesions (chancre, usually painless) will appear at the site of infection (primary syphilis), followed by an eruption on the mucous membranes and skin (secondary syphilis).⁶

Syphilis can manifest in the oral cavity in the form of chancre, mucous patches, focal epithelial hyperplasia (FEH), and papilloma/condyloma.^{3,9} Benzathine penicillin is an essential drug recommended by the WHO for the management of syphilis.⁵ FEH is a rare condition that is frequently found in immunocompromised individuals.¹⁰ This case report will discuss FEH as a rare oral manifestation in syphilis patients as a result of sexually transmitted infections in homosexual patients.

CASE

A 25-year-old homosexual man came to the Oral Medicine Department complaining of sores on his palate for 3 weeks, and 2 weeks later a painless lump appeared. He is an HIV sufferer, and he has been taking anti-retroviral (ARV) treatment since six months before. He also felt pain when opening his mouth, especially when yawning, as well as itching on the face and genital area. Extraoral, maculopapular rash were found on the palms and soles. Intraoral, there was a single mass on the palate about 2 cm in diameter, the consistency was soft, did not bleed easily, stalked, movable, and had the same color as the surrounding tissue without causing pain (Fig. 1a). There was a reddish band-like area that was suspected to be linear gingival erythema (LGE) at the maxillary anterior gingival margin (Fig. 1b).



Figure 1. The clinical condition at the first visit: a. a single mass on the palate about 2 cm in diameter, soft in consistency, did not bleed easily, stemmed and movable, and had the same color as the surrounding tissue; b. a reddish band-like area on the maxillary anterior gingival margin.

CASE MANAGEMENT

On the patient's first visit to the Oral Medicine Department, we carried out both subjective and objective examinations. On subjective examination, the patient complaining of sores on his palate for 3 weeks, and 2 weeks later a painless lump appeared. He also felt

pain when opening his mouth, especially when yawning, as well as itching on the face and genital area. Objective examination revealed on extraoral, maculopapular rash found on the palms and soles. Intraoral, there was a single mass on the palate about 2 cm in diameter, the consistency was soft, did not bleed easily, stalked, movable, and had the same color as the surrounding tissue without causing pain (Fig. 1a) also a reddish band-like area that was suspected to be linear gingival erythema (LGE) at the maxillary anterior gingival margin (Fig. 1b). We provide pharmacological therapy 0.2% chlorhexidine gluconate mouthwash three times a day to prevent secondary infections, and vaseline album cream to be applied on dry lips. In addition to pharmacological therapy, the patient was also given non-pharmacological therapy consisting of education to maintain oral health by brushing teeth and tongue with a soft-bristled toothbrush at least twice a day, after breakfast and before bedtime. Based on his sexual behavior history, we also gave him education regarding the dangers of oral sex. The management of this patient also involved other departments, the Dermatology and Venereology Department and the Periodontic Department, but we were unable to obtain photo documentation from each department. On the same day, the patient underwent laboratory examination with Hepatitis B Surface Antigen (HBsAg) immunochromatography, Venereal Disease Research Laboratory (VDRL), and Treponema Pallidum Hemagglutination Assay (TPHA) as the first treatment from the Dermatology and Venereology Department, then allowed to go home. Laboratory examination results (Fig. 2) showed HBsAg immunochromatography (negative), VDRL titer (reactive), and TPHA titer (reactive), so that the diagnosis of syphilis could be established.

Lab No.	Order Date	Order No.	Sex	DOB	Age
[REDACTED]	2023-10-03	[REDACTED]	Laki-Laki	[REDACTED]	[REDACTED]
GIGI ORAL DIAGNOSTIK					
Diagnosa Awal					
S20 SIFILIS					
Nama	Flag	Hasil	A. Satuan	Nilai Normal	
HBsAg Kompatipori 0		Non Reaktif	U	Non Reaktif	
TPHA TITER 0		Reaktif	U	Non Reaktif	
VDRL Kuantitatif		Reaktif	U	Non Reaktif	

Figure 2. Laboratory examination results

On the ninth day after the first visit, the patient came to the Periodontic Department for a scaling procedure to remove the calculus as one of the linear gingival erythema (LGE) causes. On the next day, he received Benzathine penicillin G (2.4 million single-dose units intramuscularly) from the Dermatology and Venereology Department. Unfortunately, we didn't get any documentation from those two departments due to department policy. On his second visit to the Oral Medicine Department, one month after administration of

Benzathine penicillin G, oral lesions on his palate was healed (Fig. 3a) also the scaling procedure succeeded in eliminating linear gingival erythema (Fig. 3b).



Figure 3. The clinical condition 1 month after administration of Benzathine penicillin G (dated March 10, 2022); a-b. oral lesions were healed.

DISCUSSION

Syphilis is an infectious disease caused by *Treponema pallidum* and mainly transmitted by sexual contact.^{7,11} Syphilis has become a global health problem that increasing over the last decade. This condition is mainly due to the increasing spread of the disease, especially among gays, bisexuals, and men who have sex with men (MSM).⁹ This corresponds to the patient's condition in this case report, who had same-sex sexual behavior. The course of the disease is divided into several stages, namely primary, secondary, and tertiary syphilis; oral lesions can occur at all stages.^{9,11-13} Clinical manifestations can develop from primary stage to another if the patient did not get appropriate treatment. Syphilis has the potential to cause serious complications and is closely related to HIV infection.¹¹ A person with primary or secondary syphilis who has sores or rash can transmit the disease to others. Firstly, primary lesions appear at the infected site (external genitalia, vagina, anus, or rectum). Primary lesions that do not get proper treatment will proceed to the secondary stage. Secondary syphilis develops as a result of the systemic spread of spirochetes from the site of the primary infection and affecting multiple organs. This condition is mostly manifests on the skin, mucosa, and nerves.⁹ Establishing the diagnosis of syphilis is based on the detailed anamnesis regarding to the social life of the patient and his sexual partners, the results of clinical examinations, and the results of laboratory tests. Laboratory examination using Rapid Plasma Reagent (RPR) or Venereal Disease Research Laboratory (VDRL).¹²

Oral manifestations of syphilis are conditions that are rarely recognized by the patients or the physicians. Oral lesions occur in approximately 30% of patients with secondary syphilis, although oral ulceration is rarely a major manifestation of the disease.¹² Oral manifestations of syphilis including focal epithelial hyperplasia (FEH) or fibrous hyperplasia, papilloma or condyloma, and lymphoid

hyperplasia.³ Focal epithelial hyperplasia (FEH) or Heck's disease is a rare benign lesion that can be found on the oral mucosa and usually experienced by children or adults with immunocompromised conditions.^{10,14,15} This disorder was first reported by Dr. Heck and his colleagues in 1965. The disease is more common in young people and is sometimes linked to a genetic predisposition.^{14,16} Risk factors for FEH including crowded environments, poverty, malnutrition, poor oral hygiene, and immunocompromised conditions. Globally, this condition is rare but is commonly found among Inuit and Indians living in Central, North, and South America, the Khoi-San people of South Africa, and Eskimos from Greenland and Northern Canada.^{14,17,18} FEH can also be caused by human papillomavirus (HPV) types 13 and 32 infection,^{14,19} more often in women than in men.^{15,18}

FEH can also appear as a comorbidity with other diseases. Patients with immunocompromised conditions have a higher incidence than the general population. Diseases with low levels of CD4+ T cells, such as HIV and intestinal lymphangiectasia, have been reported to be associated with FEH.^{16,18} A similar condition is also demonstrated in HIV patients who have FEH in their oral cavity.

Clinically, FEH is characterized by nodules about 1–3 cm in diameter, soft, single, or multiple, have the same color as the surrounding mucosa, the surface is elevated, can appear on the lips, buccal mucosa, labial, tongue, gingiva, and palate.^{14,18,20} The diagnosis is made based on clinical manifestations and confirmed by biopsy; however, HPV involvement is rare. This oral lesion may be a sign of HIV infection, therefore laboratory tests are carried out to confirm this condition and other sexually transmitted infections, such as syphilis.^{18,20} Management of FEH is usually unnecessary because most lesions are self-limited and do not show a tendency to become malignant.^{14,20}

In this case, the patient is an HIV-AIDS sufferer who was undergoing ARV therapy. The laboratory examinations that have been carried out include HBsAg immunochromatography, VDRL, and TPHA. HBsAg immunochromatography examination was carried out to determine the presence of the hepatitis B virus; VDRL examination was carried out to determine the appearance of antibodies against *Treponema pallidum* bacteria; and TPHA examination was carried out to determine the presence of *Treponema pallidum* bacteria. The HBsAg immunochromatography examination showed negative results, the VDRL titer was reactive, and the TPHA titer was reactive, therefore the diagnosis was syphilis. The therapy administered was intramuscular Benzathine penicillin G 2.4 million units of a single-dose which was successful to remove the mass on his palate that he complained of. The therapy is an antibiotic used to treat syphilis and prevent infections caused by other microorganisms that are sensitive to penicillin. The World Health Organization

recommends administering Benzathine penicillin G (2.4 million units) as syphilis therapy.²¹ Benzathine penicillin G is active against gram-positive bacteria, including beta-hemolytic streptococci, *Treponema pallidum* and *Treponema carateum*. Benzathine penicillin G is a bactericidal beta-lactam antimicrobial, inhibiting the cell wall peptidoglycan biosynthesis during the active multiplication stage, which will inhibit bacterial peptidoglycan transpeptidase and causing the cell wall to become unstable, resulting in lysis and bacterial cell death. Benzathine penicillin G is available as an injection administered intramuscularly.²²

Linear gingival erythema is a band-like erythematous line on gingival margin, most often affecting gingiva of the anterior teeth, but it can also be seen on the gingiva of the posterior teeth. The trigger for this condition is often associated with *Candida spp.* and is often seen in people with HIV-AIDS. The mechanism of HIV interacting with oral conditions and altering the immune response is still not clearly understood. Oral microflora such as *Candida spp.*, is mentioned to have an important role in the activation of the inflammatory response, releasing inflammatory mediators such as interleukin-1 (IL-1), tumor necrosis factor- α (TNF- α), nuclear factor-kappa B (NF- κ B), and interferon gamma, which results in the destruction of the periodontal tissue.^{23,24} Scaling was also carried out in order to treat the *linear gingival erythema* on the maxillary anterior gingival margin, and it succeeded in eliminating this condition.

The management of oral manifestations in sexually transmitted diseases such as syphilis requires cooperation both from the multidisciplinary staff and from the patients themselves. Education and support for patients undergoing treatment is very important. Dentists have an important role in dealing with oral manifestations, therefore knowledge regarding to sexually transmitted diseases such as syphilis is needed. Healing in this patient within 1 month of therapy with Benzathine penicillin G and scaling; however, relapses can still occur, that's why patient cooperation is required to comply with the education provided. Syphilis is a sexually transmitted disease caused by *Treponema pallidum*, often seen in the male population who have sex with male. This condition may manifest in the oral cavity as a focal epithelial hyperplasia. Focal epithelial hyperplasia is a rare benign lesion of the oral mucosa which is not recognized by either the patient or the physician. Knowledge of the signs and symptoms of the disease as well as a multidisciplinary approach are needed to achieve successful treatment.

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