Exclusively oral manifestation of secondary syphilis

Manifestação exclusivamente oral de sífilis secundária

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ABSTRACT

Introduction: Syphilis is at an epidemic level in the Brazilian public health and it occurs, mainly, in the genital region. However, about 5% of the syphilitic lesions are oral. **Objective:** To report a clinical case of secondary syphilis with exclusively manifestation in the oral cavity. **Case report:** A 32-year-old woman who attended the Dentistry Clinic of the Universidade Estadual do Oeste do Paraná (UNIOESTE) complaining of "sores in the mouth". Physical examination revealed a number of ulcerated, pseudomembranous and painful lesions at apex, lateral border, belly, floor and lingual frenulum. After the diagnostic hypothesis of syphilis was raised, serological tests for human immunodeficiency virus (HIV) 1, HIV-2, Venereal Disease Research Laboratory (VDRL) and complete blood count were requested. With the positivity up to the 1/32 titration of the VDRL examination, the patient was referred to the Parasitic Infectious Diseases Center of Cascavel, where a general physical examination and antibiotic therapy with benzathine penicillin were carried out. Subsequent to the drug treatment, the patient returned to the UNIOESTE Dentistry Clinic, where the total remission of the lesions was observed, confirming the diagnosis of secondary syphilis exclusively in the mouth. **Conclusion:** This report illustrates the importance of each phase of the disease, noting that in some cases the manifestations of the infection may be solely oral, with the dentist having a notable role in the diagnosis and reference to the adequate treatment, and the responsibility to control the necessary diagnostic procedures.

Keywords: syphilis; oral manifestations; sexually transmitted diseases.

RESUMO

Introdução: A sífilis encontra-se em estado epidêmico na saúde pública brasileira. Ela ocorre principalmente em região genital, todavia cerca de 5% das lesões sifilíticas são orais. **Objetivo:** Relatar um caso clínico de sífilis secundária com manifestação exclusivamente em cavidade oral. **Relato de caso:** Mulher, 32 anos, compareceu à Clínica de Odontologia da Universidade Estadual do Oeste do Paraná (UNIOESTE) com queixa de "feridas na boca". Ao exame físico, observaram-se inúmeras lesões ulceradas, pseudomembranosas e doloridas em ápice, borda lateral, ventre, assoalho e frênulo lingual. Depois de levantada a hipótese diagnóstica de sífilis, foi solicitada a realização de exames sorológicos para vírus da imunodeficiência adquirida (HIV) 1, HIV-2, Venereal Disease Research Laboratory (VDRL) e hemograma completo. Com a positividade até a titulação de 1/32 do exame VDRL, a paciente foi encaminhada ao Centro de Doenças Infecto-Parasitárias de Cascavel, sendo ali realizados exame físico geral e antibioticoterapia com penicilina benzatina. Posteriormente ao tratamento medicamentoso, a paciente retornou à Clínica de Odontologia da UNIOESTE, sendo observada a remissão total das lesões, confirmando o diagnóstico de sífilis secundária exclusivamente bucal. **Conclusão:** O relato ilustra a importância do conhecimento das características clínicas de cada fase da doença, ressaltando que em alguns casos as manifestações da infecção podem ser unicamente orais, tendo o cirurgião-dentista notável papel no diagnóstico e referência ao tratamento adequado, além da responsabilidade de dominar as manobras diagnósticas necessárias.

Palavras-chave: sífilis; manifestações orais; infecções sexualmente transmissíveis.

INTRODUCTION

Syphilis is a systemic bacterial infection caused by spirochete *Treponema pallidum*. Their means of transmission are most closely linked to the sexual contact, the vertical transmission from mother to fetus and hematogenous via, through blood transfusion, in rare cases[1].

The *T. pallidum* ethiopathogeny is related to the production of mucopolysaccharidoses that dissolve mucopolysaccharides — responsible for the union of vascular cells — , thus allowing the passage to perivascular spaces, promoting collapse, thrombosis, vascular obstruction and necrosis[2].

Chancre is the initial lesion of the infection and is characterized as an eroded and ulcerated lump, painless, with bases and hardened margins, measuring approximately 1 to 2 cm in diameter, which marks the site of the microorganism inoculation and usually manifested 21 to 30 days after contamination[3], accompanied by multiple and bilateral ganglionary reaction, non-suppurative, of hard and painless nodules[1]. It most often affects the ano-genital and oral areas[3], and most of extragenital chancres occur in the mouth (40–70%)[4].

Following primary syphilis, secondary syphilis arises along with a variety of systemic signs: pharyngitis, myalgia, arthralgia, prostration, headache and generalized lymphadenopathy[5]. Manifesting itself four to six weeks after the onset of chancre and having spontaneous remission three to 12 weeks after the onset of symptoms, secondary syphilis presents a range of extremely heterogeneous lesions, which labeled the disease as the "great copycat". Generally, the first impressions of this stage show generalized rash involving all the trunk and extremities, including injury and palmar-plantar known as syphilitic roseolas[4].

Mucosal plaques are most commonly observed in the mouth: painful, oval, serpiginous, slightly elevated erosions with erythematous borders, usually covered by a whitish membrane. They occur due to areas of intense exocytosis and spongiosis of the mucosa[3]. Another frequently observed in secondary syphilis is the canine condyloma lata, with characteristics very similar to mucous plaques, but with a nodular and firm surface[4].

Over the course of two years of infection, secondary syphilis alternates between periods of onset and remission of symptoms, until a large latency period sets in[1]. The reactivation of the disease corresponds to the tertiary stage of syphilis, which affects 30% of patients with secondary syphilis untreated, and more serious complications are registered[6]: syphilitic gum, neurosyphilis and cardiovascular syphilis[4].

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Syphilitic gum is characterized as a destructive lesion, mostly painless, nodular, which invades the mucosa underlying tissues. In the oral cavity, the hard palate is the place of greatest involvement, having palatal perforation and oral-sinus communication results of its destructive capacity[4].

In 20 years, a type of syphilis with high mortality rate was observed. From 1998 to the present day, 160,000 cases of congenital syphilis were reported in children under 1 year of age in the Brazilian territory[7]. It occurs by transplacentary transmission of the mother to the fetus at any stage of the disease, and the risk of contamination is proportional to the amount of bacteria in the maternal blood circulation. The mortality reported in the infected conceptuses is about 40%[1]. The congenital syphilis stigmas can be observed in the maxillofacial complex, and systemically. The neonate may present ectodermal alterations, mucocutaneous rash, osteochondritis, periostitis, rhinitis, destruction of the vomer bone, Hutchinson's incisors and muriform molars[3].

OBJECTIVE

To report the clinical case of secondary syphilis with exclusively oral manifestations.

CASE REPORT

Female patient D. M. U., 32 years old, leucoderma, smoker for 16 years, without extrabucal alterations, attended the Clinic of Stomatology of the Universidade Estadual do Oeste do Paraná (UNIOESTE), in the city of Cascavel, state of Paraná, with main complaint of "painful wounds in the mouth".

Intraoral examination revealed multiple ulcerated lesions, covered by whitish membranes and surrounded by a discrete erythematous halo that affected the region of the apex and dorsum of the tongue (**Figure 1**), tongue lateral border of the right and left and lingual frenulum (**Figure 2**). The complaint of painful symptomatology was evident, and the patient reported evolution of five months for the lesions. When asked about a periodic visit to the gynecologist, the patient reported having had a consultation in the six months preceding the search for care at the Clinic of Stomatology, where no alterations had been observed. There was also a report that the three years previously to the onset of the lesions were marked by the absence of sexual intercourse.

In view of the diagnostic hypothesis of syphilis mucous plaques, a request for serological tests for human immunodeficiency virus (HIV) 1, HIV-2 and Venereal Disease Research Laboratory

Figure 1 – Lesions in apex and dorsum of the tongue prior to the treatment.

Figure 2 – Ulcerations in lateral right and left border and lingual frenulum prior to treatment.

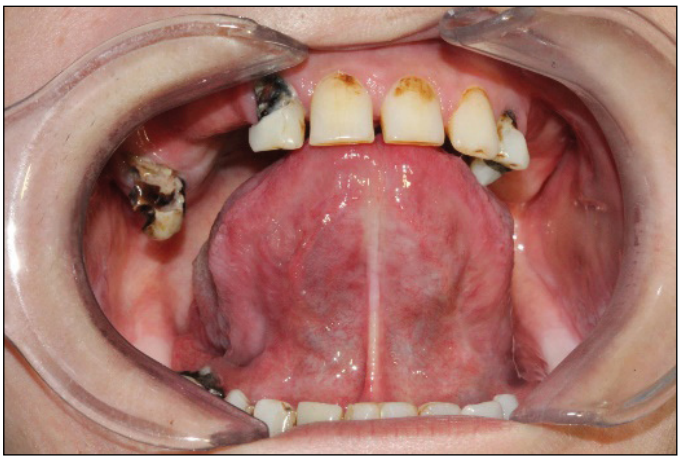

Figure 3 – Remission of lesions in lingual apex, dorsum and frenulum after treatment.

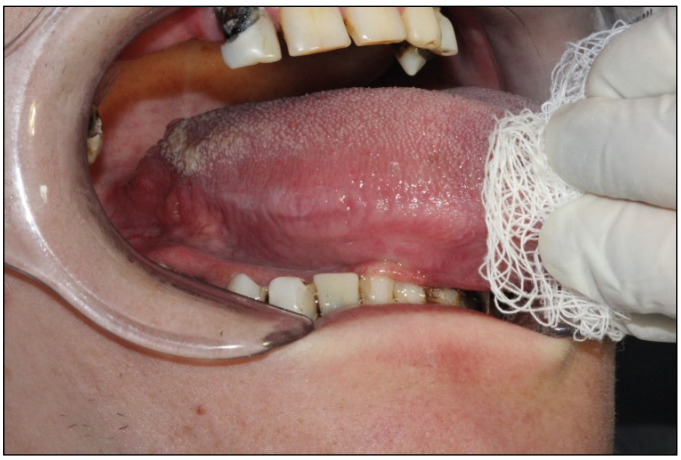

Figure 4 – Remission of lesion in lateral right tongue border after treatment.

(VDRL) was held, in addition to complete blood count. The results revealed absence of alterations in the blood count and negativity to HIV-1 and HIV-2. However, the VDRL test showed positivity up to 1/32 titration.

The patient was referred to the Specialized Center for Parasitic Infectious Diseases (Centro Especializado de Doenças Infecto-Parasitárias — CEDIP), in the city of Cascavel, where the infectology team performed a general physical examination, noting the absence of extraoral lesions and confirming the diagnosis of secondary syphilis. After antibiotic therapy with benzathine benzylpenicillin 2.4 million IU IM, the patient returned to the Clinic of Stomatology of UNIOESTE, where the total remission of all lesions was observed (**Figures 3, 4 and 5**). At the end of the treatment, a serological test was carried out showing the efficacy of antibiotic therapy and serological cure of the patient.

DISCUSSION

In the last five years, Brazil has faced increasing numbers of syphilis notification. It can be attributed to a greater readiness of access to rapid testing, reduction of condom use — largely related to improved antiviral therapy for HIV/acquired immune deficiency syndrome (AIDS) and reduced deaths from the disease — , health professionals' resistance to administering penicillin in the basic attention, worldwide shortage of the antibiotic, as well as the implementation of compulsory notification of syphilis cases[7].

Although oral manifestations of syphilis are more likely to be observed in the secondary phase, all stages of the disease may exhibit oral lesions[6]. Primary syphilis has chancre as the most characteristic injury. In secondary syphilis, mucous plaques and condyloma lata play an important role of the spectrum of lesions observed. In tertiary syphilis, syphilitic gum, with its power of destruction, the tongue becomes atrophic, lobulated, fissurated, and with leucoplastic plaques dorsally. And the oral stigma of congenital syphilis is mostly reflected in the changed morphology of incisors that exhibits a shape similar to a screwdriver and molars with cuspid projections resembling blackberries[3].

The diagnosis of syphilis is related to the investigation through non-treponemic tests — VDRL and rapid plasma reagin (RPR) — and treponemic ones, such as fluorescent treponemal antibody absorption test (FTA-ABS), microhemagglutination assay for *Treponema pallidum* (MHTP) and enzyme-linked immunosorbent assay (ELISA). Dark field microscopy, silver impregnation and cytopathology have doubtful diagnostic value and are not much recommended, since the oral cavity has other treponema species that can be mistaken with *T. pallidum*[5]. In the present case, the diagnosis was based on the clinical findings and on the VDRL reagent result, since at the end of the antibiotic therapy the lesions had total remission, and the histopathological analysis was dispensable by means of biopsy. It is worth to notice, however, the importance of this type of examination to favor additional evidences of the diagnosis of syphilis in controversial cases[4].

Figure 5 – Remission of lesion in lateral left tongue border after treatment.

With the diagnosis of secondary syphilis, questions were raised about the clinical chronology of the disease. This phase manifests from three to 12 weeks after the onset of the chancre[4], and alters periods of outbreak and remission of symptoms over the course of two years until a large period of latency is installed[1]. A possible untruth about sexual practices was speculated, since this information characterizes an embarrassing subject and allows the patient to exercise the right to preserve his/her privacy.

It is believed, however, that periods of absence of characteristic inflammatory symptoms of the infection may occur and are caused due to some particularities of *T. pallidum*. Although the bacterium expresses numerous lipoproteins that would be capable of awakening and activating defense cells, they are covered by a fragile membrane that does not have lipopolysaccharides, a highly pro-inflammatory glycolipid, capable of activating immune responses. The populations of *T. pallidum* are extremely heterogeneous, containing bacterial specimens that can bind to antibodies and others that are not capable of such immunologic activity. This set of distinct characteristics causes the spirochete to proliferate and disseminate systemically, without activating the immune system, being a microorganism with a complex capacity of "furtive pathogenicity"[8]. Thus, although three years of possible primary infection have been reported, the diagnosis of secondary syphilis was plausible.

The peculiarity of the presentation of isolated oral lesions of the disease motivated a literary survey in the MEDLINE and PubMed databases, by means of the keywords "isolated oral syphilis", "oral syphilis" and "rare oral syphilis manifestation", also seeking cases of exclusively oral syphilis. The period corresponding to the 50 recent years of scientific production was analyzed, and the existence of only five cases registered that fit in the investigated situation was found[9-13]. No information about any possible factor that provides this situation — it is only speculated that the popularization of the practice of unprotected oral sex may have enabled the increase in the incidence of oral syphilitic lesions (not necessarily as isolated manifestation)[3] — , as well as some statistical data referring to the percentage of incidence of the same clinical presentation (absence of extraoral manifestation of syphilis), was found, being the oral exclusivity considered an extremely rare characteristic of the infection[6].

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Conflict of interests

The authors declare no conflict of interests.

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