Leprosy of the hard palate: A rare case report

Sushma Bommanavar, Yashwant Ingale1, Manjusha Ingale2, Sanyukta Ingale3

Department of Oral Pathology and Microbiology, School of Dental Sciences, Krishna Institute of Medical College, Deemed to be University, Karad, 1Department of Oral Pathology and Microbiology, M. A. Rangoonwala College of Dental Science, Pune, Maharashtra, 2Dr. Ingale Institute of Dental Sciences, Pune, Maharashtra, 3BDS Student, DY Patil Dental College, Pimpri, Pune, Maharashtra, India

Abstract

Leprosy is a chronic granulomatous infection caused by Mycobacterium leprae, a bacillus that presents a peculiar tropism for the skin and peripheral nerves. Leprosy instigates various types of clinical presentation and exerts influence on the patient’s immune response. The clinical gamut of leprosy ranges from the tuberculoid form (TT) to the disseminative and progressive lepromatous form (LL). Oral lesions are uncommon but, when present, occur in the lepromatous form and are broadly divided into nonspecific and specific lesions. In this article, we present a case of leprosy of the hard palate in a 25-year-old male. The case is been presented for its rarity.

Keywords: Lepromatous leprosy, leprosy, leprotic oral lesions, Mycobacterium leprae, palate

INTRODUCTION

Leprosy, also known as Hansen’s disease is a chronic infectious granulomatous disease caused by Mycobacterium leprae, an acid-fast bacillus that has peculiar tropism for skin, mucous membranes of the nose, peripheral nerves,[1,2] and eyes.[3,4] It was discovered by the physician, Gerhard Armauer Hansen, in 1873.[1,2] The word Leprosy is derived from the Latin word “leporus,” which means “defilement.”[1] Recently, the number of cases of leprosy has decreased histrionically, from 5.2 million cases in 1985–204,800 cases at the end of 2009 as a result felicitous detection and effective multi-drug treatments.[5,6]

Leprotic oral lesions which are more common in the lepromatous form of leprosy, have a great epidemiological importance as a source of infection[6] with the prevalence reported to range from 19% to 60% of the patients.[7,8] The presence of oral lesions is directly proportional to the duration of the disease, indicating that these are a late manifestation.[7] When untreated, these lesions results in characteristic deformities, thus emphasizing the importance of mucosal lesion control.[9] In this article, we present the clinical and microscopic finding of a case of the lepromatous leprosy on the hard palate in a 25-year-old male.

CASE REPORT

A 25-year-old male presented with the complaint of progressively growing mass at the palatal region of 2 years duration. The growing mass was a size of the peanut which progressively increased to the present...
size [Figure 1]. He did complain of some difficulty in mastication and phonetic articulation but gave no complaints of the loss of sensation of the hard palate or the oral mucosa. His general and physical examination was normal, except for his suffused face. Blood investigations were requested, which rendered normal results. On intraoral examination, a large, well defined lobulated mass was seen on the hard palate. The growth was hard and was fixed to the underlying maxillary bone. The surface of the growth did not bleed on touch, and there was no associated lymphadenopathy. Keeping in mind the gradually progressive nature of the growth and the clinical appearance of the patient, we arrived at a provisional diagnosis of a lepromatous lesion on the hard palate. Incisional biopsy was performed, and a histopathological diagnosis of lepromatous leprosy was confirmed [Figures 2-4]. The patient underwent excisional biopsy under general anesthesia.

DISCUSSION

Leprosy is a disease of great antiquity and continues to be significant health problem worldwide. It is caused by an obligate, intracellular, Gram-positive, aerobic, rod-shaped carbol-fuchsin positive acid-fast bacillus, *M. leprae* that has a peculiar tropism for skin, peripheral nerves, upper respiratory tract and eyes, thus producing skin lesions, nerve degeneration, anesthesia and when untreated results in characteristic deformities. Leprotic oral lesions develop insidiously and are generally asymptomatic secondary to nasal changes. The most frequently affected intraoral site is the hard palate. The greater prevalence in men could be explained by the fact that women seek doctor’s advice earlier, perhaps for esthetical reasons.

*M. leprae* has affinity for cooler regions of the body for its multiplication. This hypothesis was confirmed by
Hastings et al. who observed that the bacterial index was much higher in the skin where the mean surface temperature was 32.5°C as compared with skin with a mean surface temperature of 33.46°C.[10] Scheepers et al.[11] found the hard palate to be the most frequent site of oral involvement in leprosy, followed by the soft palate, labial maxillary gingival, tongue, lips, buccal maxillary gingival, labial mandibular gingival, and the buccal mucosa. This was found to correlate with their mean surface temperatures. Hence, lower the mean surface temperature, higher was the frequency of involvement. About 75% cases of oral lesions seen on the anterior part of hard palate, shows a mean surface temperature of 27.4°C.[12] Based on this speculation, a pathophysiological mechanism has been suggested for the oral involvement as follows a nasal lesion with obstruction of the air flow leads to mouth breathing, which is very common in lepromatous leprosy. This causes a decrease in the intraoral temperature, mainly in sites near the air intake and in the anterior areas, thus allowing the harboring of the bacillus.[3-7]

Classification

Leprosy is a spectrum of disease classified into two basic types, i.e., tuberculoid leprosy and lepromatous leprosy.[1,13] The tuberculoid form is a localized, deforming, self-limited process with the strongest response, whereas lepromatous leprosy is relatively anergic state and when left untreated, may be fatal.[9] Between these two stages, there are several borderline forms of the disease such as borderline tuberculoid, mid-borderline, and borderline lepromatous (BL).[13]

According to the World Health Organization system, Leprosy is classified based on the number of skin lesions and proliferation of bacteria as “paucibacillary” (“pauci” refers to a low quantity) and “multibacillary” (“multi” refers to a high quantity).[1,2]

The Ridley-Jopling classification combines clinical, histopathological, and immunological criteria to identify five leprosy forms: tuberculoid (TT), borderline tuberculoid, mid-borderline (BB), BL, and Lepromatous (LL).[1,2,14]

Also according to the Indian classification, leprosy is divided into tuberculoid, borderline, lepromatous, indeterminate, and neurotic forms.[14]

Several different approaches were also adopted by the World Health Organization (WHO) such as SHAY scale that provides five gradations and ICD-10.[2]

Pathophysiology

Human cell biology central dogma contains antioxidant defense mechanism such as Vitamins A, C, and E enzymes such as superoxide dismutase and catalase. These function to perk up the generation of reactive oxygen species (ROS) and hence provide protection against the oxidative damage to cells. The “antioxidant hypothesis” propounded that antioxidants such as Vitamin C, Vitamin E, carotenoids present in fruits and fresh leafy vegetables afford protection against the oxidative damage to cells thus act as scavenging free radicals in the aqueous and lipid phase of cells. The major defense system in pathogenesis of leprosy is the macrophage system, and its enigmatic mechanism is as shown in the flowchart:

1. Once infected through respiratory droplet infection, the foamy macrophages in skin and nerve show increased phagocytosis, enzyme activity and oxygen consumption known as respiratory burst but they are unable to kill the M. leprae
2. Respiratory burst activity leads to production of a variety of molecules and free radicals called ROS such as superoxide anion, hydrogen peroxide, and hydroxyl radicals. These ROS can damage polyunsaturated fatty acids, proteins and nucleic acids
3. Increased state of lipid peroxidation due to inadequate scavenging of ROS due to decreased level of antioxidant defense such as Vitamin E and Vitamin C (as leprosy patients are mostly from low socioeconomic status, they do not get antioxidant-rich diet such as fruits and fresh leafy vegetables every day) leading to increased oxidative stress
4. This oxidative stress which occurs due to derangement in the balance between ROS and natural antioxidants may be responsible for the clinical manifestations seen in patients of leprosy.[6]

Thus, it is hypothesized that enhanced lipid peroxidation observed in leprosy patients can also be attributed to a large extent to the insufficiency of vitamins in the diet.[6]

Lepra reaction

Is a hypersensitivity reaction due to the presence of the pathogen in tissues that can occur during the course of the disease.[13]

Types

Tuberculoid type

Involvement of skin, nerve, and regional lymph nodes. Lesions are early, intermediate, and developed types. Early lesions show hypopigmented macules whereas intermediate lesions demonstrate larger with elevated margins. Developed lesions are densely anesthetic with
loss of sweat glands and neuritic pain. Loss of eyebrows and eyelashes are also noticed.

**Borderline type**
Similar to tuberculoid type except the lesions are larger and more in number and lepromatous type except less anesthetic and less in number.

**Lepromatous type**
Involvement of skin, nerves, mucous membrane, lymph nodes, eyes, skeleton, testis, and other internal organs. Thickening of the skin on face can cause deeping of the natural line on the forehead and other parts of face giving an Leonine face appearance. Earlobes are also thickened. Additional features include ocular damage and facial palsy.

**Indeterminate type**
Hypopigmented macules usually seen on covered areas of body and on face.

**Oral manifestation**
Intraorally, the lesions vary from enanthemas to ulcers, perforations and scars, passing through papules, nodules (lepromas), and superficial erosions.

Palatal lesions-demonstrate infiltration, ulceration, perforation, and reddish or yellow-reddish nodules, sessile or pedunculated, varying from 2 to 10 mm, some confluent, and prone to ulceration.

Tongue lesions, especially on dorsal surface of anterior two-thirds demonstrate superficial erosions with loss of the papillae and longitudinal fissures with nodular infiltration that could lead to a “Paving stone appearance.” Scarring can also occur.

Uvula-extensive fibrosis in extreme cases with partial or complete destruction is seen. Lips – Macrostomia or macrocheilia are seen. Gums Chronic periodontitis especially behind the maxillary central incisors are seen and are often continuous with the lesions on hard palate.

Histological stained section reveals infiltration of macrophages, lymphocytes and plasma cells with an abundance of acid-fast bacilli prominently in oral lesions of lepromatous leprosy. Scheepers et al. described epithelial atrophy to be a feature seen only in patients with lepromatous leprosy. Abreu et al.[15] studied 19 patients with multibacillary leprosy between 2000 and 2002, and concluded that clinical alterations in the oral mucosa do not imply disease involvement and should be confirmed by histopathological examination. Hyperkeratosis is seen in few cases. Grenz zone, which is a consistent feature in skin lesions, is rarely seen in oral lesions.[14]

Diagnosis of leprosy continues to depend predominantly on clinical criteria, with laboratory findings usually being auxiliary in nature. The presence of two out of three cardinal features are considered to be diagnostic, and these include decreased or loss of sensation in skin lesions or the peripheral nerve involved, enlargement of the peripheral nerve, and presence of acid-fast bacilli in smears.[14] The method of choice, as propagated by the WHO, is the slit-skin smear.[9] Various assays have been developed for a demonstration of *M. leprae* – specific DNA and ribosomal RNA sequences in various specimens such as nasal smears, skin, and blood. Polymerase chain reaction (PCR) for bacterial DNA is most sensitive and can detect fewer than 10 organisms. The majority of patients with suspected leprosy who have negative screening smears have positive PCR results. With early diagnosis and treatment, progression of disease can be limited, but recovery of neuronal function is variable. Even when disease is controlled, the stigma and social isolation often persist.[8]

**CONCLUSION**
Leprotic oral lesions are more common in the lepromatous form of leprosy, and hence the oral cavity should be examined carefully when leprosy is been suspected. When untreated, these lesions results in characteristic deformities, thus emphasizing the importance of mucosal lesion control and its early detection so that individual suffering from such infection can lead productive lives in the present community.

**Declaration of patient consent**
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**
There are no conflicts of interest.

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