International Journal of Research in Medical Sciences
Gonnuru VK et al. Int J Res Med Sci. 2023 May;11(5):1844-1847
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20231371

Case Report

Rare occurrence of palatal perforation in a case of leprosy: a case report and literature review

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Received: 17 March 2023
Accepted: 15 April 2023

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ABSTRACT

The aim of the study was to demonstrate the rare occurrence of coexistent nasal septal and palatal perforations in a case of lepromatous leprosy. Nasal septal perforation is a full-thickness defect of the nasal septum which includes the bilateral mucoperichondrial flaps and structural middle layer. It occurs mostly on the anterior cartilaginous septum. Most of the patients are asymptomatic and the two-third of the people affected show no nasal complaints. Palatal perforation is said to have occurred when there is a development of communication between bilateral nasal cavities and the oral cavity. Whereas in our case, septal perforation was found to be co-existent with palatal perforation which is a rare condition. A 32-year-old, otherwise healthy patient presented with complaints of nasal regurgitation of food contents and shortness of breath while eating for 3 months. Septal mucosal biopsy, biopsy from the palatal perforation and the left ear nodule was taken without any complications, and tissue was sent for histopathological examination which showed features suggestive of lepromatous leprosy. The occurrence of coexistent septal and palatal perforation is rare and it is even rarer to be found in a case of lepromatous leprosy. Hence proper history should be elicited and adequate investigations such as histopathological examination should be performed in cases of coexistent nasal septal and palatal perforation.

Keywords: Hard palate, Perforation, Lepromatous leprosy

INTRODUCTION

Leprosy, also known as Hansen's disease, is a chronic granulomatous infection caused by the pathogenic bacterium *Mycobacterium leprae*. This disease is found to affect both sexes with a higher prevalence in males.

The mode of transmission is via droplets. It can affect the nerves, skin, eyes, and nasal mucosa leading to different manifestations.[1] If it affects the nerves, it leads to loss of touch and pain sensations which when left untreated may lead to paralysis.

Leprosy affecting the skin results in discolored patches and nodules. If the bacteria affect nasal mucosa, it leads to a stuffy nose and nasal bleeding. Involvement of the oral cavity is rarely seen in leprosy and when present is found in lepromatous leprosy cases. The prevalence of oral cavity involvement is found to be 19 to 60%. The oral lesions are varied and include infiltration, perforation, ulceration, and reddish-yellow nodules.[2] The presence of oral lesions is a late manifestation of leprosy.[3]

Leprosy is a curable disease and if diagnosed early and treatment initiated, disability can be prevented.

CASE REPORT

A 32-year-old male presented to the ENT outpatient department with complaints of regurgitation of food through the nose and shortness of breath for 3 months. The patient was asymptomatic 3 months ago when he noticed nasal regurgitation of food contents. He also complained of shortness of breath while eating. He gave no history of fever, cough, cold or vomiting. He was found to have no known co-morbidities. On inquiry about addictions, the patient was found to be an alcoholic. On examination, vitals were found to be stable. Systemic examination was normal.

The skin over the nose was found to have a hypopigmented macule. Nasal cavity examination revealed nasal crusts and perforation involving septal cartilage. Oral cavity examination revealed the presence of an oval perforation measuring about 1×0.5 cm in the hard palate resulting in the formation of an oronasal fistula (Figure 1). The patient was planned for a biopsy from the perforation margins for histopathological examination. The pre-op period was uneventful. The patient was shifted to the operation theatre and a biopsy was taken from the margins of the palatal perforation, septal perforation and the left ear nodule under local anesthesia without any complications.

Hemostasis was secured and closure was done with 3-0 catgut sutures. The postoperative period was uneventful. The tissue was sent for histopathological examination. Microscopic examination revealed sheets of foamy histiocytes containing bacilli in the cytoplasm, plasma cells and a few eosinophils in the dermis (Figure 2). Connective tissue stroma was also found to contain foamy histiocytes. AFB positivity was seen with 5+ globi (Figure 3). These features were suggestive of lepromatous leprosy.

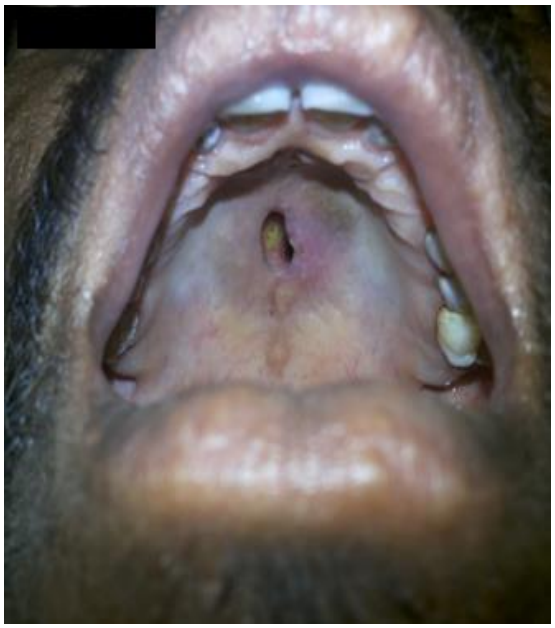

Figure 1: Perforation in the hard palate.

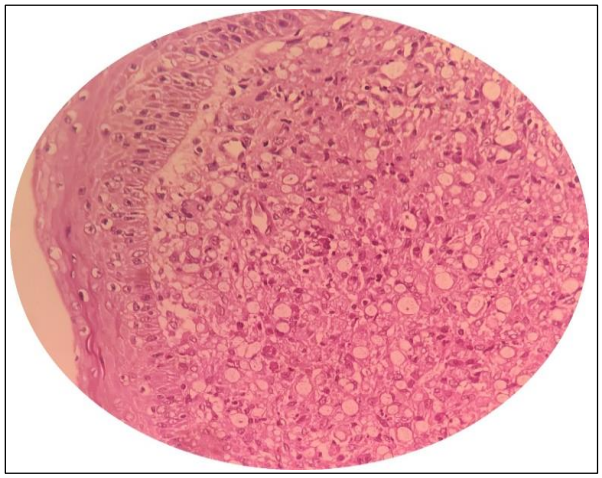

Figure 2: Histological picture showing foamy histiocytes in the dermis.

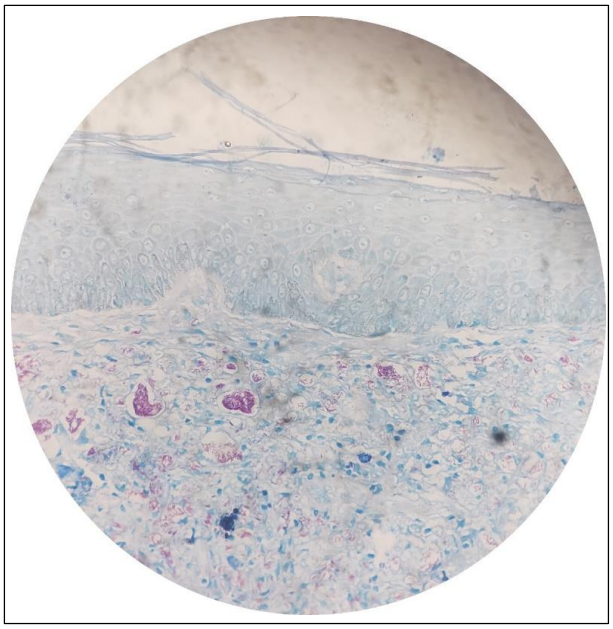

Figure 3: AFB positivity seen on histo-pathological examination with globi.

DISCUSSION

The palate forms the roof of the mouth and is divided into hard palate (anterior four-fifth) and soft palate (posterior one-fifth). The hard palate serves as a partition between the nasal and oral cavities and is formed by the palatine processes of maxillae in the anterior two-thirds and horizontal plates of palatine bone in the posterior one-third.[4] There can be several potential causes of palatal perforation as listed further. It could be developmental due to the failure of integration of palatal shelves in the prenatal period. Infections like leprosy, tuberculosis, tertiary syphilis, rhinoscleroma, naso-oral blastomycosis, leishmaniasis, actinomycosis, histoplasmosis, coccidioidomycosis and diphtheria may cause palatal perforation. Autoimmune conditions such as systemic lupus erythematosus, Crohn's disease, sarcoidosis and

Wegener's granulomatosis may result in perforation of the palate.

Tumors extending from the maxillary sinus or nasal cavity may spread to the palate causing perforation. Illicit drug abuse of cocaine, heroin and narcotics is a well-known cause of perforation. It could be iatrogenic sometimes encountered following a tooth extraction. Some rare causes like rhinolith may also cause perforation.[5] Hence these differentials should be kept in mind when a case of palatal perforation is seen. The treatment is dependent on the diagnosis and thus a detailed history, physical examination, and laboratory studies should be done to narrow down the differential diagnosis.

Leprosy is a chronic granulomatous disease caused by the bacteria *Mycobacterium leprae*. The routes of transmission are not yet conclusive and one of the prime factors involved is nasal droplet infection. Leprosy has been found to primarily affect the nerves, skin, eyes, and nasal mucosa. Involvement of the oral cavity is rare in leprosy and is usually often found in individuals belonging to the Virchowian group. The prevalence of oral lesions was found to range from 19% to 60% among cases of lepromatous leprosy.[6] *Mycobacterium leprae* is reported to have a higher affinity for cooler regions of the body. This hypothesis was confirmed by the observation that the bacterial index was higher in the skin with lower mean surface temperature as compared to a higher mean surface temperature.[3] The palate is a structure found to be crossed by both the nasal and the oral air currents resulting in its temperature being maintained about 1-2°C below the body temperature.[6] The most frequent site of involvement in the oral cavity in cases of leprosy is the hard palate. The frequency of involvement of other oral sites was found to directly correlate with the mean surface temperature at these sites.[3]

Pathophysiology

A pathophysiological mechanism for oral involvement has been suggested. Nasal lesions lead to obstruction of airflow which promotes mouth breathing. This results in decrease in intraoral temperature in the anterior areas near sites of air intake. This favours multiplication of bacilli and thus the oral lesions.[3]

Classification

Leprosy is broadly classified into two types: tuberculoid type and lepromatous type.

According to Ridley-Jopling classification which is a combination of clinical, histopathological, and immunological criteria, leprosy has been divided into five forms: tuberculoid leprosy (TT), borderline tuberculoid (BT), mid-borderline (BB), borderline lepromatous leprosy (BL), and lepromatous leprosy (LL).[3] According to the WHO system which employs the number of skin lesions and proliferation of bacteria as the basis, leprosy is classified into ‘paucibacillary’ and ‘multibacillary’.[3]

Tuberculoid leprosy

Lesions of tuberculoid leprosy are usually seen involving the skin and peripheral nerves. The skin lesions may be hypopigmented macules or plaques which are well-demarcated, dry, scaly, anhydrotic and hypesthetic with erythematous or raised borders. Peripheral nerve involvement is usually asymmetrical and associated with hypesthesia and myopathy. The bacilli are absent or few on microscopic examination.[7]

Lepromatous leprosy

Lesions of lepromatous leprosy are usually symmetrical. The skin lesions can be nodules or plaques. Diffuse dermal infiltration may be seen in few cases. These lesions when present on the face result in characteristic facies called the *Leonine facies*. Manifestations that develop later in the disease process include madarosis, pendulous earlobes and dry scaling skin. Nerve involvement in these cases is insidious and more extensive when compared to tuberculoid leprosy and the resultant nerve enlargement is usually symmetrical. The bacilli are numerous and found in large clumps called globi on microscopic examination.[7]

Oral manifestations

The oral lesions are usually asymptomatic.[8] Oral cavity lesions may occur even in the absence of visible lesions in other parts of the body. It involves lips, tongue, hard and soft palate, buccal mucosa, uvula, and gums.

The hard palate is the most commonly involved site in the oral cavity due to the factors mentioned above. The commonly found lesions include erythematous papules. These lesions increase in size and number and coalesce leading to formation of a submucosal infiltrate. Progression of the disease results in loss of mucosal shininess and ultimately gives rise to a matt-like appearance. Progression of disease results in palatal perforation leading to development of symptoms like dysphagia.[8]

Involvement of the tongue results in superficial erosions, loss of papillae, nodular infiltration, and longitudinal fissures. This gives rise to a paving stone appearance which is seen on examination of the tongue. The dorsal surface of the anterior two-thirds of the tongue has been reported to be most commonly involved by leprosy.[3] Extensive fibrosis is seen in the uvula and in extreme cases partial or complete destruction may develop.[3]

Involvement of lips may present as either macrostomia or macrocheilia.[3] Leprosy can affect the gums, especially behind the upper central incisors resulting in lesions of chronic periodontitis which is often found to be in continuity with the lesions on the hard palate.[3]

Diagnosis

Diagnosis of leprosy depends on clinical criteria and laboratory findings are auxiliary. The clinical criteria include hypesthesia in the skin lesions or area of innervation of involved peripheral nerves, enlargement of involved peripheral nerves and acid fast bacilli in slit skin smears. The diagnostic method of choice is a slit skin smear. Tissue diagnosis such as biopsy from the local site is confirmatory of diagnosis. Other methods include polymerase chain reaction (PCR) to detect bacterial DNA.[3]

Lepromin test

The lepromin skin test measures the cellular response to lepromin. It provides information regarding the ability of host T cells to respond to *Mycobacterium leprae* and the chances of granuloma formation. A negative lepromin test is seen in LL or BL leprosy cases and indicates the lack of a protective cellular response.[9]

Treatment

The WHO recommended multidrug therapy is followed for the treatment of leprosy. This regimen consists of a combination of two or three of the following drugs: rifampicin, dapsone, and clofazimine. The regimen and duration of therapy depend on the WHO classification of leprosy. In multibacillary leprosy, a combination of rifampicin 600 mg monthly+dapsone 100 mg+clofazimine 300 mg monthly and 50 mg daily is given for 12 months. In paucibacillary leprosy, a combination of rifampicin 600 mg monthly+dapsone 100 mg daily is given for 6 months.[9]

Treatment of specific oral lesions should be done only after completing the regimen for eradication of the bacterial disease. In our case, where palatal perforation was present as a manifestation of leprosy, repair of the palatal perforation should be done after eradication of the bacterial load. Repair of palatal perforation is done by palatoplasty.

CONCLUSION

The oral cavity must be examined in a patient with leprosy since lesions of the oral cavity may be missed and are potential sources for the spread of the disease. Oral cavity lesions may occur in leprosy even in the absence of visible lesions in other parts of the body. Histopathological examination of the tissues serves as a good diagnostic means in such doubtful cases. Serious complications may occur in lesions of the palate when compared to other parts of the body. Hence early detection and treatment of leprosy helps prevent the spread of the disease and prevents the development of disability.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Gonnuru VK, Dandala SR, Albi I, Abbugari R, Bhaskar J, Varma V. Rare occurrence of palatal perforation in a case of leprosy: a case report and literature review. Int J Res Med Sci 2023;11:1844-7.