

Case Report

Extrapulmonary Tuberculosis Involving Parotid and Ramus Region: A Forgotten Diagnosis

Vanaja Reddy¹, Arati Chaudhary², P.V. Wanjari³, Ashish M. Warhekar⁴, Sweety Lalawat⁵, Mimansha Verma⁶

^{1,4}Reader, ²Professor, ³Professor and Head, ^{5,6}Post Graduate student,
Department of Oral Medicine & Radiology, Modern Dental College and Research Centre, Gandhi Nagar, Indore

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ABSTRACT

Tuberculosis is a major health problem in developing nations. Extra-pulmonary form of this disease is uncommon, but its prevalence has increased in the last couple of years. This can affect any organ system and involvement of head and neck region is our prime concern.

Involvement of parotid gland is rare and presents as localized glandular swelling giving a non-specific clinical picture. Since a broad spectrum of pathological conditions affecting salivary gland may mimic such presentation, the possibility of tuberculous involvement of gland is often overlooked.

This case report highlights that despite of its rare occurrence, tubercular parotid lymphadenitis should be considered in differential diagnosis of a discrete parotid gland swelling/mass.

Introduction

Tuberculosis (T.B) is a prevalent systemic bacterial infectious disease frequently caused by mycobacterium tuberculosis complex. This disease poses a major health hazard and is of a concern worldwide, particularly in developing countries. India has the highest burden of T.B in the world, with an annual incidence 2 million cases. Majority of the patients have latent infection rather than active form of T.B. Therefore it is not uncommon for the patient to be asymptomatic.^{1,2}

The extra-pulmonary form of T.B accounts for about 10-15% of the cases and lymphadenopathy is its common manifestation. However apart from cervical lymphadenitis, T.B affecting head & neck region is rare accounting for only 1% of the cases, therefore is often skipped as a possible diagnosis from the dental clinician's viewpoint, resulting in inappropriate or delayed treatment and related complications.^{3,4}

This case report emphasizes that although rare, the possibility of tuberculosis infections causing lesions & swellings in oro-facial region should be considered amongst the differential diagnosis and necessary investigation should be carried out for confirmation followed by referral for appropriate medical treatment.

Case Report

A 25 year old female patient reported to the Dept. of Oral Medicine & Radiology, with swelling in left middle and lower third of face since 6 months. The swelling was gradual in onset, associated with intermittent and dull aching type of pain.

Past medical and family history of the patient was non-contributory. There was no history of chronic cough or weight loss. General physical examination revealed that she was ill-built, malnourished and afebrile.

* Corresponding author: Dr. Sweety Lalawat, P.G student, Dept. Of Oral Medicine & Radiology, Modern Dental College And Research Centre, Gandhi Nagar, Indore, (M.P) Email: sweetylalawat@yahoo.in, Mobile: 09826831031

Extra-oral examination revealed a diffuse swelling in left pre-auricular region measuring approx. 4×4 cms in size. Overlying skin of the swelling appeared to be stretched with no evidence of any secondary changes. Another similar type of swelling was present in left submandibular region measuring approximately 3×2 cms in size consistent with enlarged submandibular lymph nodes. (fig. 1)

On palpation, swelling was mildly tender, firm in consistency, non-fluctuant, compressible, non-reducible, temperature of overlying skin was not raised. There was no evidence of trismus and facial nerve function was normal.

Intra-oral findings were non-contributory.

Therefore on the basis of history and clinical examination of swelling in pre-auricular region differential diagnosis of the following were considered: benign tumor of parotid gland, chronic sialadenitis and tubercular lymphadenitis.

The routine blood investigations were found to be normal. Screening test for HIV, HbsAg, HCV was negative. ESR was raised to 85 mm/hour. Mantoux test was positive.

Panoramic radiograph revealed a single, well-defined, non-corticated radiolucency on left middle 1/3rd of ramus, measuring approx. 2×2 cm in size. (fig. 2) Chest X-ray revealed normal findings.

Ultrasonography revealed multiple enlarged lymph-nodes in intra-parotid, pre-auricular, sub-mandibular & posterior triangle lymph node region. These nodes showed increased vascularity on Doppler USG suggestive of infective etiology. There was heterogenous collection in left deep parotid space with ecogenic foci within with suspected bony erosion. (fig. 3)

USG guided FNAC from left submandibular lymph-node revealed granulomatous reaction comprising of



Fig. 1: Two diffuse swellings; i) In left preauricular region; ii) In left sub- mandibular region

lymphocytes, plasma cells, macrophages & epithelioid cells with caseous necrosis which was highly suggestive of tubercular granulomatous reaction.

Final diagnosis of extra-pulmonary tuberculosis was made.

Patient was referred to tuberculosis center and standard anti-tubercular regimen was instituted for 6 months followed by which extra-oral features were restored to normal. The radiolucent area within the ramus of mandible also showed evidence of bone healing. (fig. 4) The patient is under regular follow-up.

Discussion

Tuberculosis is a form of chronic necrotizing granulomatous disease and has wide spread distribution and variable clinical presentations. Its extra-pulmonary form can virtually affect any organ system with head and neck region being our prime concern. ^{2,5}

Lymph node involvement is most common form of extra-pulmonary tuberculosis and in head and neck region, the most common site being cervical lymph nodes which clinically manifests as tubercular lymphadenopathy. Amongst cervical group of nodes, anterior cervical nodes are frequently involved followed



Fig. 2: A single, well-defined, non-corticated radiolucency on left middle 1/3rd of ramus, measuring approx. 2×2 cm in size.

by posterior cervical, submandibular and supraclavicular nodes respectively.⁶

In our case posterior cervical, submandibular lymph nodes were involved. This involvement may occur during primary tuberculous infection or secondary form as a result of reactivation of dormant foci or direct extension from a contiguous focus.

Other sites of tubercular involvement in head and neck region, which are the rare sites includes flat bones like mandible and salivary glands. These are involved in the present case, making it a rare case of extra-pulmonary tuberculosis. The probable common routes of transmission to these sites which appears to be apt for our case include, hematogeneous spread and lymphatic spread. Apart from these certain specific routes for salivary gland includes ascent of infection through salivary gland ductal system and in case of bone regional extension from overlying soft tissues.⁷

Extra-pulmonary tuberculosis involving jaw bones manifests as an uncommon form of chronic osteomyelitis which causes slow necrosis and may involve the entire jaw bone. Mandible is more frequently involved than maxilla with greater affinity for alveolar and angle regions. But in our case ramus region of mandible was

involved, making it a rare entity of extra-pulmonary tuberculosis.

The destruction of the bone in radiographs appears as blurring of trabecular details with irregular areas of radiolucency in initial stages. Later there is erosion of the cortex followed by gradual replacement of bone by soft tuberculous granulation tissue with little tendency to repair.²

In our case, erosion of cortex was seen which showed evidence of healing on follow-up radiographs. This denotes that if the lesion is detected early, the disease can be completely curable and can lead to reversal of all destructive bony changes.

Another site involved in our case was salivary glands, a rare entity even in countries with high incidence of tuberculosis such as India. The probable cause for this rarity is because of inhibitory effect of salivary secretion on mycobacterium.

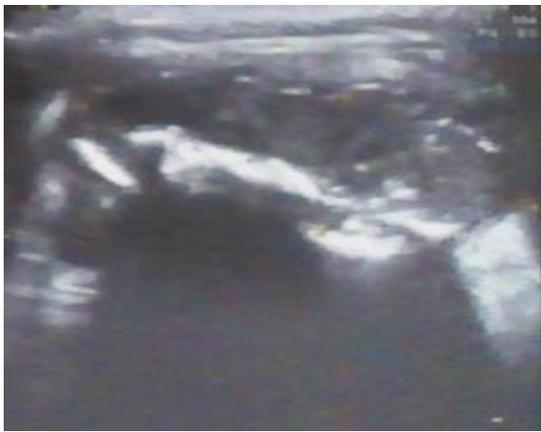
Tubercular involvement of the salivary glands is commonly seen secondary to systemic dissemination of pulmonary tuberculosis than as primary extra-pulmonary tuberculosis. Parotids are involved 70% of the time, frequently due to localized tubercular disease, as seen in



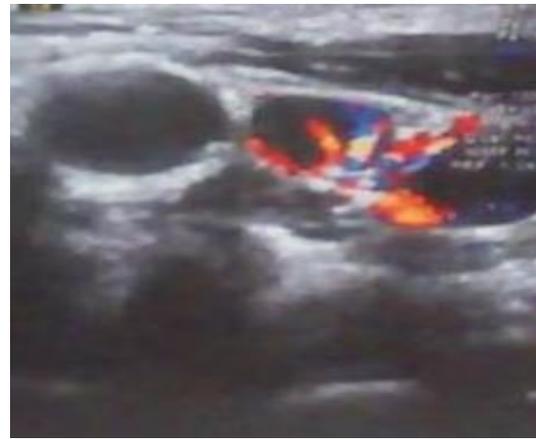
(i)



(ii)



(iii)



(iv)

Fig. 3: i) Multiple enlarged lymph-nodes in left submandibular region.
 ii) Multiple enlarged lymph-nodes in left pre-auricular region.
 iii) Nodes showed increased vascularity on Doppler USG
 iv) Heterogenous collection in left deep parotid space with ecogenic foci within with suspected bony erosion.

our case. It clinically manifests either as, diffuse parenchymatous involvement which is less common or as tubercular parotid lymphadenitis which shows pericapsular or intraparotid nodal involvement. The later form of manifestation is seen in our case. This nodal involvement is characterized by localized glandular swelling with gradual enlargement and without general symptoms.^{7,8}

The clinical picture is usually non-specific and many other broad spectrum of pathological conditions affecting parotid gland may mimic parotid T.B lymphadenitis. These may include:

I . Infection or inflammatory conditions of salivary glands like:

Sialadenitis: This cause sudden painful enlargement of salivary gland secondary to bacterial infection and the acute form of this will show signs of acute inflammation

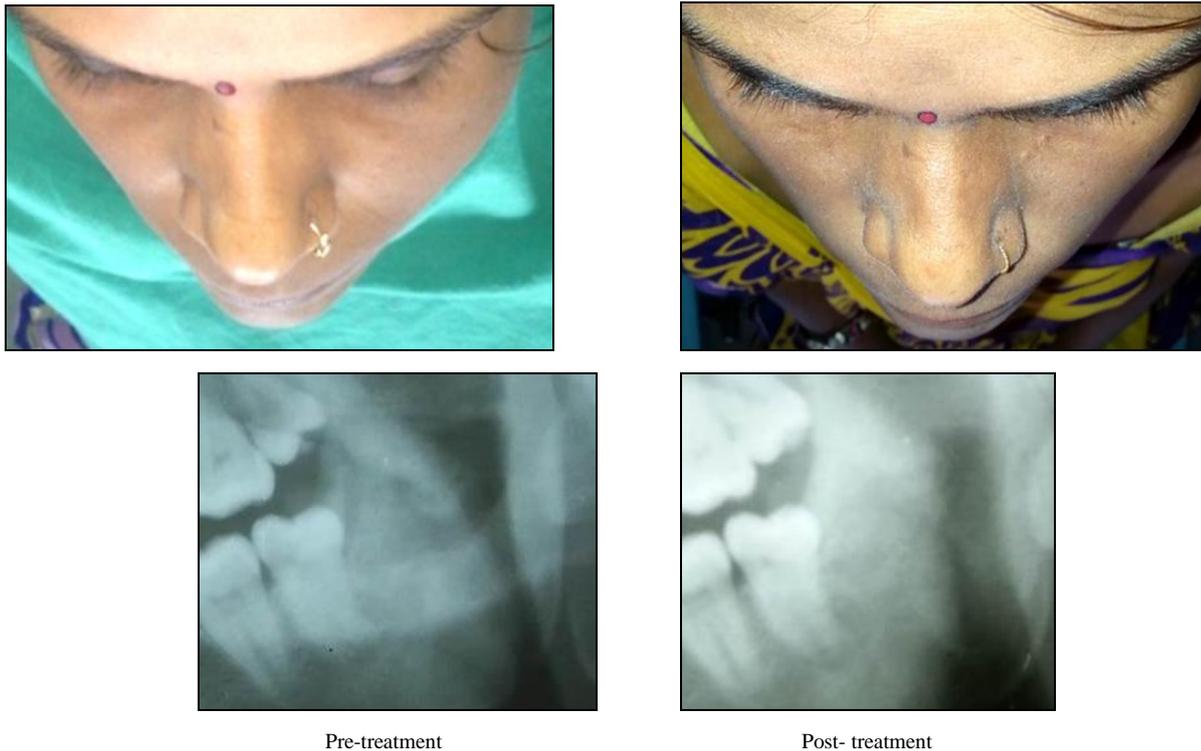


Fig. 4: Follow-up after 6 months; i) Extra-oral features were restored to normal; ii) Radiolucent area within the ramus of mandible also showed evidence of bone healing.

and purulent discharge from duct opening upon milking of gland. Chronic form is characterized clinically by intermittent, often painful swelling.

Viral parotitis (Mumps): This has an acute onset and parotid gland is involved primarily, usually bilaterally (75%).^{9,10,11}

These diffuse inflammatory conditions of the gland are often difficult to differentiate clinically from tubercular involvement gland if culture of secretions from Stensen's duct or saliva is negative for acid fast bacilli.

HIV-SGD: Primary sign of HIV-SGD is salivary gland swelling, primarily in the parotid glands and frequently bilateral. It is associated with a diffuse infiltrative CD8+ cell lymphocytosis.

Benign lymphoepithelial lesion/cysts commonly develop later in the course of the disease. Ultrasonography demonstrates large anechoic/hypoechoic areas.

II. Salivary gland tumors like Pleomorphic adenoma and Muco-epidermoid carcinoma. These present as painless enlarging swelling of the gland. Ear lobe will be raised in such cases. Tuberculosis of the parotid gland presents with difficulties in diagnosis because of the similarity of the clinical presentation to that of a neoplasm. Also certain imaging findings are indistinguishable from that of a neoplasm and therefore it requires histopathological confirmation.

III. Granulomatous diseases of gland like Sarcoidosis causes usually bilateral painless parotid enlargement. This may be multinodular or less commonly diffuse. Histological findings are characteristic and reveal non-caseating granulomas with epithelioid cell proliferation.^{9,10,11}

IV. Others:

Mikulicz's disease: This is an immune condition characterized by symmetric lacrimal, parotid, and submandibular gland enlargement with associated lymphocytic infiltrations.

Lymphoma: Majority of major salivary gland lymphomas are Non-Hodgkin's lymphoma of B-cell lineage and will therefore show microscopic picture of B-cell lymphoma tissue.

Kimura's disease: An immune-mediated inflammatory disease characterized by a triad of painless subcutaneous masses in the head or neck region, blood and tissue eosinophilia, and markedly elevated serum immunoglobulin E levels.^{9,10,11}

Various investigation procedures are used for the diagnosis of tuberculosis. These include: Chest x-ray, Mantoux test, Zeil- Neelsen staining, PCR, Sputum test and Biopsy.²

Certain imaging modalities like ultrasonography, certainly has some role in salivary gland swellings and in evaluation of lymph nodes. It demonstrates lymph nodes as per their nodal distribution, size, shape, internal architecture. Tubercular lymph nodes appear as hypoechoic structures with absent hilum, usually round in shape with unsharp borders. Matting and perinodal edema are certain additional findings. Doppler USG further provides details of vascularity.¹²

Further CT scan & MRI are useful for defining the extent of lesion & detect any concomitant deeper lesions and

osseous involvement. The findings on these modalities are highly sensitive.⁵

Recently, USG guided FNAC have emerged as an acceptable, safe alternative to biopsy with high specificity and sensitivity in cases of salivary gland swellings and lymph nodes.⁵

Cytological examination reveals epithelioid cells, granulomatous areas with lymphoid cells and caseous necrosis.¹³

In our case also USG guided FNAC was conclusive and yielded the diagnosis.

Conclusion

Multidrug resistant T.B is spreading its tentacles and affecting individuals based on their immune conditions. Taking into consideration the occurrence of these cases dentists should be vigilant and incorporate extrapulmonary tuberculosis in differential diagnosis of head & neck swellings. This awareness not only helps in diagnostic process but also in choosing the rightful investigation, prevention and treatment options.

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