DOI: 10.7860/JCDR/2016/17195.7501

Case Report

Dentistry Section

# Primary Paranasal Tuberculosis in a Diabetic Mimicking Odontogenic Infection: A Rare Case; A Unique Presentation

AMIT GUPTA[1], MONICA MEHENDIRRATTA[2], CHANCHAL SAREEN[3], ANJU AGGARWAL[4]

## ABSTRACT

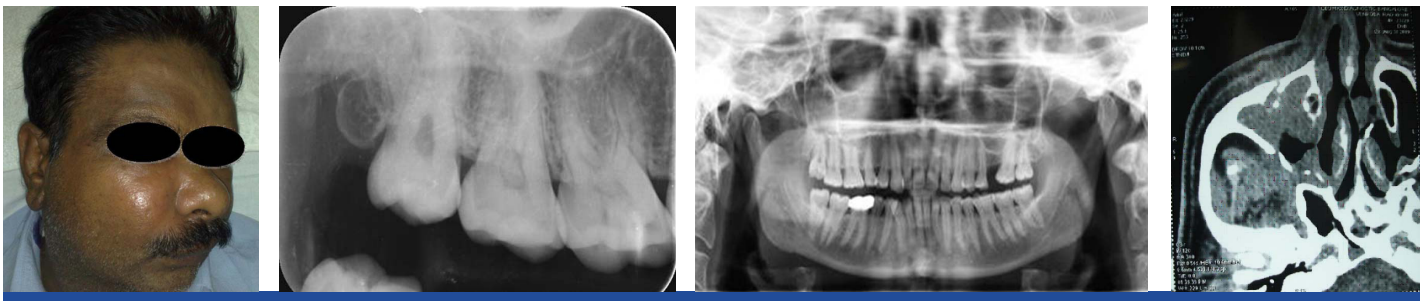
The incidence of Tuberculosis (TB) is high especially in developing countries but primary para-nasal TB is still a rarity. The latter often remains quiescent until it reaches an advanced stage and offers a diagnostic challenge. In the present case report maxillary sinus TB mimicked a destructive periodontitis induced space infection, thus causing a delay in treatment. The present case report describes clinical presentation, diagnosis, management and outcome of a 50-year-old diabetic/HIV seronegative patient with histopathologically confirmed case of maxillary sinus TB.

**Keywords:** Chronic infection, Diabetes, Differential diagnosis, Extra-pulmonary, Fungal infection, Maxillary sinus

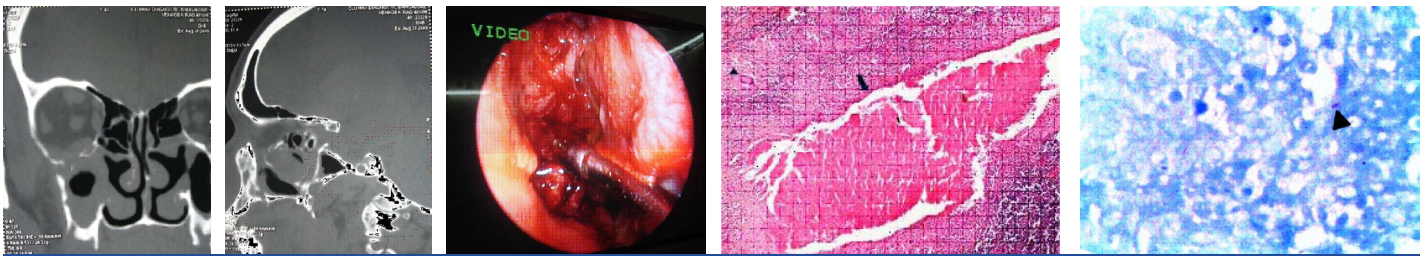
## CASE REPORT

A 50-year-old male patient reported to the Hospital out-patient department, Lady Hardinge Medical College, New Delhi with a complaint of swelling of the face and painful, mobile upper right posterior teeth since 3 weeks. Patient had a long history of Type II diabetes with fluctuating glycaemic control in spite of medication. On examination, a diffuse swelling was present over the right peri-orbital area, infra orbital region extending up to the upper lip and right alar margin and posteriorly up to the right tragus of the ear [Table/Fig-1]. The tissues were indurated on palpation with fluctuation elicited in the right maxillary buccal sulcus in the region of the right maxillary molars. The right maxillary second and third molars were tender and intra-oral periapical radiograph revealed destructive periodontitis [Table/Fig-2]. A provisional diagnosis of the right buccal and canine space infection secondary to periodontitis involving the maxillary molars was made. Hospital admission and routine hematological investigations were carried out, with administration of Empirical antibiotics. An incision and drainage through an intra-oral buccal sulcus approach to drain the contents and extraction of the mobile maxillary second and third molars was planned and performed under local anaesthesia. Scanty purulent pus was drained and sent for microbiological stains, culture and sensitivity testing which yielded negative growth after 72 hours.

Patient was discharged on the third postoperative day with relief from pain and mild resolution of the facial swelling, and was advised to continue the course of antibiotics for a period of one week. On follow up after a week, the extraction and incision sites exhibited uneventful healing. However, the extra-oral facial swelling persisted and patient complained of post nasal discharge and blockade of the right nasal passage and pain in the para nasal sinus on the right side. An orthopantomograph was done which revealed a hazy right maxillary sinus suggestive of chronic inflammatory right maxillary sinusitis [Table/Fig-3]. Diagnostic Magnetic Resonance Imaging (MRI) was performed to determine the source and extent of the persistent facial soft tissue swelling, which revealed mucosal thickening with polyposis involving the right maxillary, frontal, ethmoidal and sphenoidal sinuses with destruction of anterior and posterior walls of the right maxillary sinus and the hard palate, with



**[Table/Fig-1]:** Profile picture showing clinical presentation pre-operatively. **[Table/Fig-2]:** IOPA revealing destructive periodontitis around maxillary molars. **[Table/Fig-3]:** Right maxillary sinus revealed cloudy appearance. **[Table/Fig-4]:** Axial CT Scan section showing involvement of right maxillary sinus.




**[Table/Fig-5]:** Coronal CT Scan section showing involvement of right maxillary sinus and presence of a soft tissue mass. **[Table/Fig-6]:** Sagittal CT Scan section showing involvement of ethmoidal and frontal sinuses. **[Table/Fig-7]:** Endoscopic view of granulomatous tissue in right maxillary sinus. **[Table/Fig-8]:** H&E stained section shows central area of caseous necrosis (arrow) enveloped by lymphocytic rim and a bipolar langhans giant cell (arrowhead). **[Table/Fig-9]:** ZN stained section showing magenta colored acid fast bacilli in right maxillary sinus (arrow head).

involvement of right orbit, infra temporal fossa and superior orbital fissure [Table/Fig-4-6].

A differential diagnosis of a fungal infection or malignancy involving the right maxillary sinus was made. Through a diagnostic trans-nasal endoscopic approach, the right maxillary sinus was viewed which revealed a brownish granulomatous mass [Table/Fig-7]. A biopsy specimen was obtained through the endoscopic approach and was sent for histo-pathological evaluation in the department of Oral and Maxillofacial pathology.

The sections revealed moderately dense stroma containing numerous ill-formed granulomas composed of central areas of caseous necrosis surrounded by a rim of lymphocytes and plasma cells including epitheloid histiocytes, langhans' and foreign body type of giant cells [Table/Fig-8]. A Ziel Nielsen stain was done on the histopathologic section which revealed the presence of few acid fast bacilli confirming the diagnosis of tubercular involvement of the right maxillary sinus [Table/Fig-9].

This was followed immediately by a routine posterior-anterior (PA) view chest radiograph and AFB staining of sputum which showed no evidence of pulmonary involvement. Right submandibular lymph node was enlarged, mobile, palpable and tender which was resolved after extraction of upper molars and empirical antibiotic therapy. Patient was then started on standard anti-TB treatment (ATT) according to Category1 with drug combination which include 2 month intensive therapy of Rifampicin, Pyrazinamide, Ethambutol and Isoniazid (H3R3Z3E3) followed by four month continual therapy (H3R3). Patient was reviewed after a period of one month when there was total resolution of the facial swelling, clearance of the nasal blockade and absence of any post-nasal discharge.

## DISCUSSION

Tuberculosis (TB) is one of the chronic diseases affecting humans known from the age of Hippocrates [1]. Pulmonary TB is the most common presentation and its prevalence is more widespread in third world countries when compared to developed nations. However, primary extra-pulmonary TB especially one involving para-nasal sinuses is rare in occurrence even in developing countries [2]. The clinical presentation of para-nasal TB as chronic non-specific suppuration of maxillary sinus can lead to misdiagnosis of odontogenic/ non-odontogenic infections which is common amongst oral and maxillofacial practitioners. The former infections are irresponsive to conventional medical line of management and thus, other disorders such as those due to a fungal infection or a malignancy involving the sinus are suspected; TB sinusitis however is frequently not assumed.

The pulmonary TB and diabetes mellitus has known relationship for years. Diabetes mellitus being an important risk factor for TB affects its course and the response to treatment. An immunocompromised host offers a fertile soil for the spread of *Mycobacterium Tuberculosis*. Eventually in diabetics, TB worsens the glycaemic control and glucose tolerance. Although such links between the extra-pulmonary TB and diabetes mellitus is not well known [3].

To the best of our knowledge the incidence of extra pulmonary TB affecting the para-nasal sinus has been remarkably low with only 20 cases reported by 1907 [2] and 40 cases being reported until 1977 [4-6] in the English language literature. Chronic inflammatory disease of the maxillary sinus is quite common and we report a unique case of primary TB of the maxillary sinus in a diabetic patient that presented clinically itself as an odontogenic infection.

The WHO's (2003) recommended definition of extra pulmonary TB states this to be a TB of organs other than the lungs e.g. pleura, lymph nodes, abdomen, genitourinary tract, skin, joints and bones, meninges, etc. Disease should be based on one culture positive specimen, or histological or strong clinical evidence consistent with active extra-pulmonary tuberculosis, followed by a decision by a clinician to treat with a full course of anti-tuberculosis chemotherapy [7].

TB is a known infectious disease caused by an intra-cellular acid-fast bacillus, demonstrated by various acid-fast stains like Kinyoun and the traditional Ziel-Nielsen staining methods [8].

Extra-pulmonary sites of TB represent just 15% of all sites [9]. Most cases of tubercular involvement of the maxillary sinus are secondary to pulmonary TB [10]. Para-nasal TB involving the maxillary sinus usually occurs directly via infected micro-droplets from nose, teeth or eye or may be by blood stream from a distant focus [11]. The contagious nature of TB of the paranasal sinuses, specifically of the maxillary sinus is the same as that of primary TB [12]. It occurs more commonly in adults and often presents with nasal discharge/ stuffiness, crusting, and occasionally, epistaxis [11].

Clinicopathologically, three types of sinonasal TB have been described in the literature: (i) submucosal involvement leading to formation of polyps, mucosal thickening, pale and boggy appearance with minimal pus discharge; (ii) rare, aggressive with bony involvement/ erosion, fistula formation and abundant discharge of acid-fast bacilli (AFB); and (iii) rare, presents with hyperplastic changes, that is, formation of granuloma, "Tuberculoma" [13]. Patient was chronic diabetic. Diabetes mellitus is not associated with extrapulmonary TB but it is associated with increased risk of active, culture-confirmed pulmonary TB [14]. There are fewer evidences related to the higher incidence of extrapulmonary TB in a diabetic patient. Thus, the relationship between the two still remains unsettled [15].

The clinical symptoms of TB may not manifest themselves until the disease is well on its way. Prognosis depends on the extent of involvement of the lesion and response to the proper treatment initiated at the right time. All of the para-nasal types TBs have a propensity to be symptom-free until the lesion has reached an advanced stage. Common symptoms that the patient often presents are nasal obstruction and rhinorrhea; however, these symptoms are non-specific, hence do not lead to a suspicion of sinus TB.

In such suspected cases of chronic maxillary sinus infection that presents as an odontogenic infection non responsive to treatment, fungal infections and malignancies should also be considered.

Allergic/ chronic fungal rhino-sinusitis is often caused by *Aspergillus flavus* spp. Allergic/ chronic fungal rhinosinusitis show allergic mucin with noninvasive hyphae, elevated total IgE, cell mediated response or both. Also, the peripheral eosinophilia, major histocompatibility complex (MHC) class II antigens and a favourable response to steroids help in determining the aetiology. Histopatholgically, the sinus lining can show microabscesses with hyphae to long standing allergic mucin with scattered hyphae, graulomatous inflammatory reaction but no invasion or necrosis. The latter is often a case or allergic fungal rhinosinusitis [16].

*Rhinosporidium seeberi* should also be considered in the differential diagnosis [17]. Rhinosporidosis produces polypoid growth with yellow pin point spots. Histopathologically, they present as eosinophilic concentric large round structures of 50-100mm in a granulomatous inflammatory tissue [16].

On the other hand deep fungal infections e.g. Histoplasmosis, Coccidioidomycosis, Sporotrichosis and Blastomycosis are characterized with occasional multinucleated giant cells. Special stains like Periodic Schiff's and Methanamine silver can help rule out the fungal elements [16].

Whereas, malignancies are often suspected or can co-exist in the third type of paranasal sinus TB. The hyperplastic type of TB can show Tuberculoma formation or present as a fluctuant swelling thus may resemble a malignancy or Potts puffy tumour respectively [18]. There are various malignancies which can take origin from sinus and can be easily differentiated through histopathological examination e.g., Squamous cell carcinoma, Non-keratinizing carcinomas, Malignancies of neuroendocrinal or glandular origin etc.

The diagnosis of TB can be made using a standard Mantoux test, bacteriological examination/culture and a histo-pathological examination that can help making a final diagnosis by revealing epitheloid and giant cells suggestive of TB. Caseous necrosis is not a characteristic feature but helps ruling out differential diagnosis which present with non caseating granulomas e.g. Wegener's granulomatosis or sarcoidosis.

Treatment is medical in nature, with Anti-Tubercular treatment regimes, combinations that include Rifampicin (450mg), Ethambutol (800mg), Isoniazid (300mg) and Pyrazinamide (750mg). Surgical treatment of TB of the maxillary sinus is not warranted as the medical line of management using anti-tubercular treatment is generally favourable.

## CONCLUSION

It is a unique case of TB of the maxillary sinus that mimicked an odontogenic infection in its clinical presentation. The clinical presentations of the patient lead to a diagnosis of an odontogenic infection arising from the right maxillary second and third molars. Conventional imaging studies revealed haziness in the right maxillary sinus, prompting us to further investigate with a Magnetic Resonance Image (MRI) which surfaced fungal sinusitis or malignancy involving the right maxillary, ethmoidal and sphenoidal sinuses with destruction of the walls of the right maxillary sinus, superior orbital fissure and the hard palate. Biopsy of the lesion was done under general anaesthesia by functional endoscopic sinus surgery. Presence of numerous, clothed and necrotic granulomas histopathologically confirmed the diagnosis of TB maxillary sinus. There was no evidence of pulmonary TB on further investigation like negative AFB staining of sputum and absence of other constitutional symptoms of cough and weight loss. Hence, this was the primary TB of the right maxillary, ethmoid and sphenoidal sinuses that was successfully managed by Anti-tubercular therapy.

## REFERENCES


[1] Oxford text book of Medicine Diabete Lemmitus 3rd edition: 2, 11;Pg:1492-1493.
[2] Gleitsmann JW. Tuberculosis of Accessory Sinuses of the Nose. *Laryngoscope*. 1907;17:445.
[3] Dooley KE, Chiasson RE. Tuberculosis and diabetes mellitus: convergence of two epidemics. *Lancet Infect Dis*. 2009;9(12):737-46.
[4] Kukreja HK, Sacha BS, Joshi KC. Tuberculosis of Maxillary Sinus. *Indian Journal of Otolaryngology*. 1977;29:27.
[5] Page JR, Jash DK. Tuberculosis of the nose and paranasal sinuses. *J Laryngol Otol*. 1974;88:579-83.
[6] Jain MR, Chundawat HS, Batra V. Tuberculosis of the maxillary antrum and of the orbit. *Indian J Ophthalmol*. 1979;27:18-20.
[7] Nissapatorn V, Kuppusamy I, Jamaiah I, Fong MY, Rohela M, Anuar AK. Tuberculosis in diabetic patients: a clinical perspective. *Southeast Asian J Trop Med Public Health*. 2005;35(4):213-20.
[8] Zhao D, Yang XM, Chen QY, Zhang XS, Guo CJ, Che XY. A modified acid-fast staining method for rapid detection of Mycobacterium tuberculosis. *J Microbiol Methods*. 2012;91(1):128-32.
[9] Penfold CN, Revington PJ. A review of 23 patients with TB: A forgotten entity. *The Laryngoscope*. 1996;34:508-10.
[10] Ravi M, Ashu G. Tubercular Osteomyelitis of maxilla. *Calicut Med J*. 2005;3(3):e4.
[11] Chawla RK, Yadav SPS, Raj B, Goyal H, Rao NV, Chawla J, et al. Tubercular Sinusitis-A Forgotten Entity. *Ind J Tub*. 1989;36:187-88.
[12] Nitassi S, Nazih N, Boujemaoui M, Essakali L, Kzadri M. Tuberculosis of the paranasal sinuses: a case report. *Fr ORL*. 2007;93:347-49.
[13] Gleitsmann JW. Tuberculosis of the accessory sinuses of the nose. *Laryngoscope* 1907;17(6):445-50.
[14] Leung CC, Lam TH, Chan WM, Yew WW, Ho KS, Leung GM, et al. Diabetic control and risk of tuberculosis: a cohort study. *Am J Epidemiol*. 2008;167(12):1486-94.
[15] Perez-Guzman C, Vargas MH, Arellano-Macias MR, Hernandez-Cobos S, Garcia-Ituarte AZ, Serna-Vela FJ. Clinical and epidemiological features of extrapulmonary tuberculosis in a high incidence region. *Salud Publica Mex*. 2014;56:189-96.
[16] Guarner J, Brandt ME. Histopathologic Diagnosis of Fungal Infections in the 21st Century. *Clin Microbiol Rev*. 2011;24(2):247-80.
[17] Chakrabarti A, Das A, Panda NK. Overview of fungal rhinosinusitis. *Indian J Otolaryngol Head Neck Surg*. 2004;56(4):251-58.
[18] Kant S, Srivastava R, Verma AK, Singh HP, Singh S, Ranganath TG, et al. Maxillary sinus tuberculosis: various presentations. *Indian J Chest Dis Allied Sci*. 2013;55(3):175-77.



**PARTICULARS OF CONTRIBUTORS:**
1. Senior Resident, Department of Oral & Maxillofacial Surgeon, Lady Hardinge Medical College & Hospital, New Delhi, India.
2. Reader, Department of Oral & Maxillofacial Pathology, ITS Dental College, Greater Noida, Uttar Pardesh, India.
3. Senior Lecturer, Oral & Maxillofacial Pathology, ITS Dental College, Greater Noida, Uttar Pardesh, India.
4. Senior Lecturer, Department of Prosthodontics, ITS Dental College, Greater Noida, Uttar Pardesh, India.

**NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:**
Dr. Amit Gupta,
H No- 4/1388, Street No-7, Bholanath Nagar, Shalimar Park, Shahdara, Delhi- 110032, India.
E-mail: aquadramit@yahoo.in



**FINANCIAL OR OTHER COMPETING INTERESTS:** None.

Date of Submission: **Oct 16, 2015**
Date of Peer Review: **Dec 21, 2015**
Date of Acceptance: **Jan 11, 2016**
Date of Publishing: **Mar 01, 2016**