

CASE REPORT

Extrapulmonary tuberculosis: atypical presentation in otorhinolaryngology

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SUMMARY

Tuberculosis (TB) continues to be a major health burden globally more so in low/middle-income countries like India. There is an increase in the prevalence of extrapulmonary TB (EPTB) because of HIV epidemics and increased usage of immunomodulating drugs. EPTB constitutes 15%–20% of all patients with TB and >50% of HIV-TB coinfecting patients. We present three such atypical presentations of EPTB in head and neck region. EPTB can mimic any disease, hence knowledge of the unusual presentations helps in making early diagnosis and thereby reduces the morbidity and mortality involved with the disease.

BACKGROUND

New surveillance and survey data from India have shown that the epidemic of tuberculosis (TB) is larger than what was previously estimated. Though there is a decrease in the incidence of TB globally, TB continues to be one of the top 10 causes of death globally.¹

TB involving any organ other than lungs is diagnosed as extrapulmonary TB (EPTB). Head and neck TB constitutes 10% of all EPTB.² We report three such cases of EPTB with unusual presentation in immunocompetent individuals.

CASE PRESENTATION**Case 1**

A 41-year-old male patient presented to ear-nose-throat outpatient department with a history of

left-sided facial swelling for 8 months, which was painless and progressively increasing in size. His family history was not significant. On anterior rhinoscopy there was purulent discharge in the left nasal cavity. The rest of the otolaryngological and systemic examination was normal.

Case 2

A 22-year-old female patient presented with a history of left-sided painless neck swelling for 2 months. On examination, the patient had an ulceroproliferative growth involving upper pole of left tonsil (figure 1A) and palpable firm jugulodigastric lymph node (figure 1B). Working diagnosis of tonsillar malignancy was done and the patient was investigated accordingly.

Case 3

A 26-year-old male patient presented with a history of swelling in the right cheek which was progressively increasing in size for 1 month. On examination, there was a bilobed cystic mass measuring 4×2 cm both below and above the right zygomatic arch (figure 2). Skin over the swelling was normal.

INVESTIGATIONS**Case 1**

Contrast-enhanced CT of paranasal sinuses showed heterogeneously enhancing mass occupying most of the left maxillary sinus with destruction of all the walls of maxillary sinus except medial wall (figure 3A,B). Mass had extended anteriorly up to muscular plane of cheek, posteriorly was invading left lateral pterygoid muscle, superiorly the inferior wall of orbit was eroded and inferiorly left superior alveolar arch was eroded. Chest X-ray was normal.

Histopathological examination of the tissue biopsy from the maxillary mass showed areas of dense lymphoid aggregates, caseous necrosis (figure 3C) and necrotising granuloma with epithelioid cells and Langhans cells (figure 3D), which led us to the diagnosis of maxillary sinus TB.

Case 2

Mantoux test was strongly positive (figure 4A).

Fine needle aspiration of left jugulodigastric node and histopathological examination of left tonsil showed necrotising granulomas (figure 4B,C).

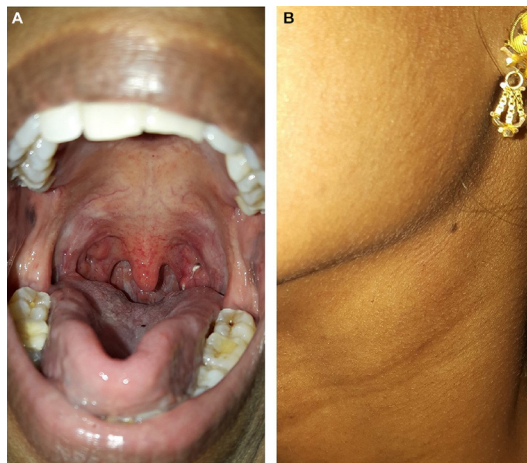


Figure 1 (A) Ulceroproliferative growth involving upper pole of left tonsil. (B) Left firm jugulodigastric lymph node.



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Figure 2 Bilobed cystic mass below and above the right zygomatic arch.

Case 3

Contrast-enhanced MRI of head and neck showed a peripherally enhancing bilobed cystic lesion in the right masseter muscle with extension up to the right zygomatic arch (figure 5A,B).

Immunological serology for *Echinococcus* was negative.

Fine needle aspiration cytology of swelling showed epithelioid macrophages in a necrotic background and smear was positive for acid fast bacilli.

DIFFERENTIAL DIAGNOSIS

Case 1

Extensive erosion of the walls of maxillary sinus with normal chest X-ray and negative Mantoux test made us think in terms of malignancy of paranasal sinus.

Case 2

Firm level 2 lymph node with proliferative growth involving the tonsil led us to the suspicion of tonsillar malignancy.

Case 3

Based on the MRI findings differential diagnosis of hydatid cysticercosis was made.

TREATMENT

All three patients were started on antitubercular therapy as per the revised national TB control programme—isoniazid 600mg, pyrazinamide 1500mg, rifampicin 450mg three doses per week for the first 2 months, followed by isoniazid and rifampicin for the next 4 months.

OUTCOME AND FOLLOW-UP

The patients were followed up throughout the therapy for 6 months and there is complete regression of the swellings (figure 5C).

DISCUSSION

The incidence of TB globally in 2015 was estimated as 10.4million. Sixty per cent of these cases accounted from six countries—India, Indonesia, China, Nigeria, Pakistan and South Africa.¹ End TB strategy endorsed by WHO targets 90% reduction in TB deaths and 80% reduction in TB incidence by 2030, compared with 2015. The success of the programme depends on early diagnosis and treatment. Atypical presentation of EPTB causes a diagnostic challenge and thereby can delay the initiation of treatment. Delay in EPTB is two or three times more than the delay in pulmonary TB.³

TB lymphadenitis is the most common presentation of EPTB. Other manifestations which can be frequently seen are in the form of disseminated disease, involving pleura, skeletal system, abdomen, cutaneous and renal system. TB of head and neck is a rare entity.

TB of nose and paranasal sinuses was first described in 1761 by Morgagni.⁴ TB can affect any sinus but the most susceptible ones are the maxillary and ethmoid.⁵ TB involving the nose and sinus is rare due to protective function of nose such as ciliary movement, bactericidal secretions and filtering action of the vibrissae.⁶ The disease is usually secondary to pulmonary TB. Extrapulmonary sinonasal TB can occur by haematogenous spread or by direct extension into the sinus.

Three types of sinonasal TB have been described:

- a. Infection is confined to mucosa only, leading to thickening of mucosa and formation of polyps. Discharge is minimal

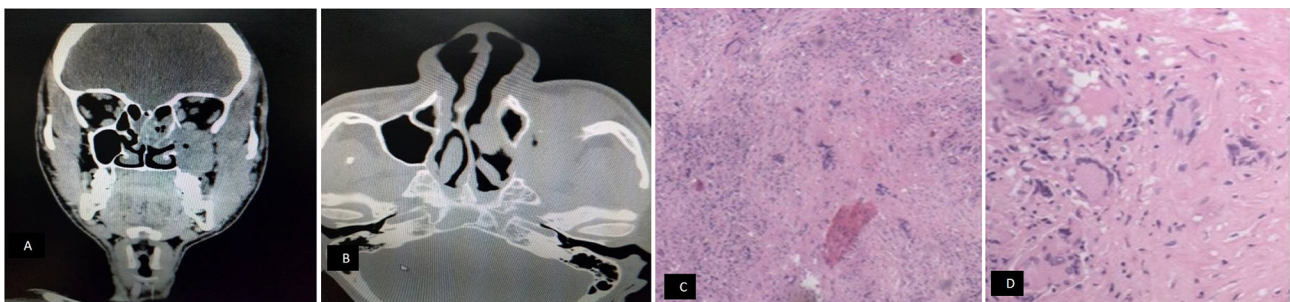


Figure 3 (A, B) Contrast-enhanced CT (CECT) of paranasal sinuses (PNS) showing mass occupying the left maxillary sinus with destruction of all the walls except the medial wall. (C, D) High-power view showing ill-defined granuloma composed of epithelioid cells, Langhans-type giant cells, lymphocytes and plasma cells.

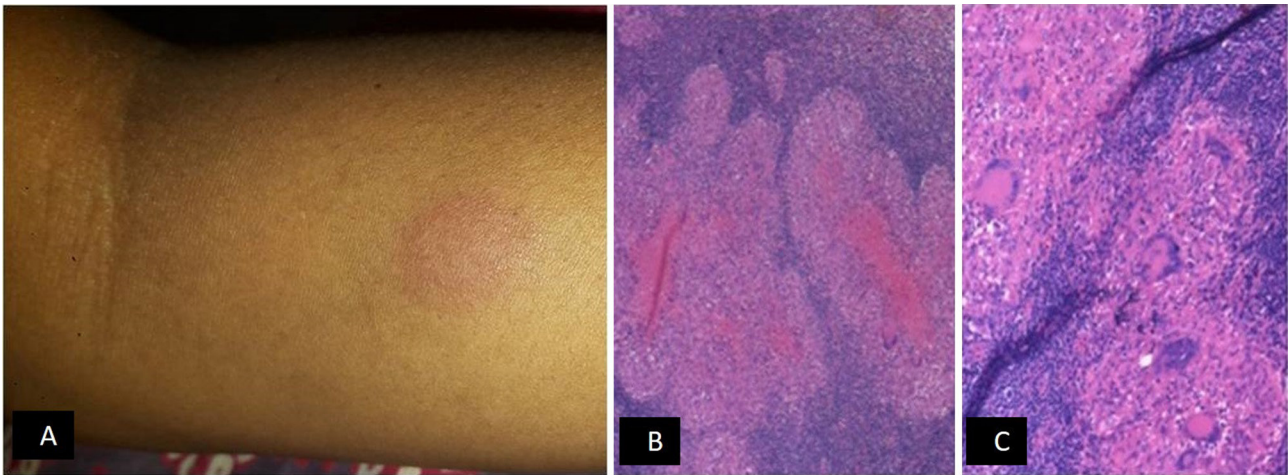


Figure 4 (A) Positive Mantoux test. (B, C) Low-power view showing well-defined granulomas composed of epithelioid cells, Langhans-type giant cells with dense infiltration by lymphocyte and plasma cells.

and making a diagnosis is difficult. This is the most common type.

- b. Bony involvement with fistula formation. Discharge is abundant with acid fast bacilli.
- c. Hyperplastic type with tuberculoma. This is the rarest type and can mimic malignancy.⁷

The initial symptoms of sinonasal TB are nasal obstruction, purulent nasal discharge and epistaxis. In case of erosion of floor of orbit they can present with epiphora and visual disturbances.⁸

Oral TB is a rare entity with tongue and palate being the most common. Though tonsil is a lymphoid organ and situated at a site which is frequently in contact with infected sputum, the incidence of tonsillar TB is less than 5%.⁹ This may be due to the

antiseptic and cleansing action of saliva, presence of saprophytes preventing the colonisation and thick stratified epithelium lining the tonsil.¹⁰ Poor oral hygiene, dental extraction and leukoplakia have been postulated to be predisposing factors for oral TB. Inhalation and colonisation of Waldeyer's ring may be one of the routes of acquiring primary TB.¹¹ Persisting sore throat, painful deglutition and unequal enlargement of tonsils with cervical lymphadenopathy should rise a suspicion of tonsillar TB.

Cutaneous TB is rare and accounts for 1%–1.5% of EPTB. Based on the pathogenesis, clinical features and histopathology cutaneous TB is classified as:

- a. Inoculation TB from an exogenous source through a broken skin in a person who was previously not infected with TB.

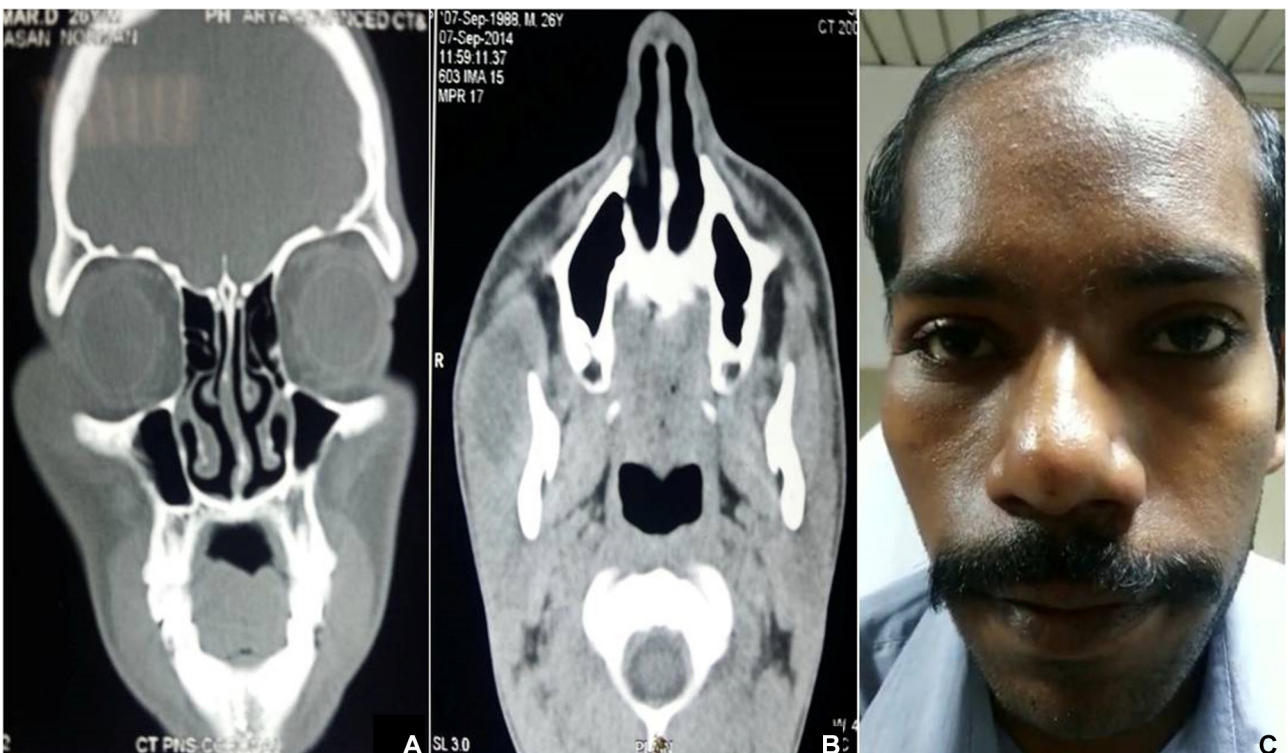


Figure 5 (A, B) Contrast-enhanced MRI showing bilobed cystic lesion in the right masseter muscle with extension up to zygomatic arch. (C) Regression of the swelling following antitubercular treatment.

- b. TB from an endogenous source such as lymph nodes, bones and joints.
- c. Haematogenous TB.¹²

TB can involve the soft tissue surrounding the bone and lead to the formation of cold abscess. However, some of abscesses have been reported without the involvement of underlying bone.¹³ Primary EPTB involving cheek is rare and confirming the site of origin of the infection is extremely difficult.

Learning points

- ▶ Incidence of extrapulmonary tuberculosis has been markedly increased due to increase in the immunodeficiency states. However, in all our three cases patients were immunocompetent.
- ▶ Tuberculosis is a great imitator and knowledge about the varied presentations of this disease helps in reducing the diagnostic dilemma.
- ▶ High degree of suspicion is required to make early diagnosis and to control the spread of tuberculosis in the society.

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