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Case Report

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Rare presentation of extra-pulmonary tuberculosis of the cheek

Prasanna Kumar Saravanam[1], Urvashi Singh[2]

1 *Associate Professor, Department of ENT, Head & Neck Surgery, Sri Ramachandra Medical College and Research Institute, Porur, Chennai-600116, India*

2 *Resident, Department of ENT, Head & Neck Surgery, Sri Ramachandra Medical College and Research Institute, Porur, Chennai-600116, India*

Abstract

Extra- pulmonary tuberculosis (EPTB) has become a rising health problem aided by a pandemic of Human Immunodeficiency Virus infection (HIV). It occurs in about 15%-20% of HIV negative immunocompetent individuals and a greater propensity to occur in those with HIV infection. It predominantly affects the lymphatics and other organ systems like – central nervous system, skeletal, pleural etc. Tuberculosis of the cheek is uncommon, and is usually associated with a concomitant oral lesion. Extra-oral Tuberculosis in the absence of an oral lesion, is a unique occurrence. Hence, it can pose a diagnostic challenge especially in a young immunocompetent individual. We therefore report a rare presentation and management of Extra-pulmonary Tuberculosis of the Cheek in an immunocompetent 26 year old male.

Keywords: Extra-pulmonary, Tuberculosis, Cheek, Immunocompetent, HIV negative.

INTRODUCTION

Tuberculosis remains a global health problem even more than a century after Robert Koch identified *Mycobacterium* as the single causative agent. Despite 1.3 million deaths (including 320,000 in HIV positive individuals) in 2012 alone, the incidence and mortality of pulmonary tuberculosis has been on the decline [1]. Though there is a declining trend of pulmonary tuberculosis, the incidence of extra-pulmonary tuberculosis is on the rise, especially in HIV positive individuals, due to the tendency of developing a disseminated disease in a background of compromised immune status. Extra-pulmonary tuberculosis, occurs with an incidence of 15%-20% in immunocompetent [2] individuals, and about 50% of HIV positive individuals [3-4]. 10% of EPTB involves the head and neck region [5], with about 0.05%-5% involving the oral region [6]. Tuberculosis of the cheek, be it primary or secondary, is an uncommon presentation especially in the absence of oral tuberculous ulcer, sinus or fistula [7].

CASE REPORT

A 26 year old male, presented to the Department of Otorhinolaryngology, Sri Ramachandra University, for evaluation of a unilateral gradually progressive swelling of the right cheek since the last one month. He gave no history of recent trauma, dental extraction, nasal discharge / nasal obstruction or fever. He had no significant past medical history (previous history of tuberculosis or exposure to tuberculosis) or family history.

He was afebrile on presenation with Pulse Rate 88/min regular in rate and rhythm, and Blood Pressure of 100/60mmHg. On clinical examination, a 4cm x 2cm swelling was present over the right cheek extending 4 cm inferolateral to the dorsum of the nose; it was a bilobed mass palpable both above and below the right zygomatic arch. The skin over the swelling was found to be normal, with no scar, sinus, fistula or ulceration. The swelling was non tender, no loco-regional change in temperature, cystic in consistency, and fluctuant with ill-defined margins. (figure 1) Lips and oral cavity examination was unremarkable; there were no lesions in the oral cavity. Nasal examination was unremarkable. Eye examination showed no chemosis or periorbital edema. Oculomotor functions were found to be intact

Complete blood count was within normal limits with Hb 12.6 g /dl and Total Count of 7200 cells /cumm. His Differential Count was as follows – polymorphs 75.9%, lymphocytes 16.7%, eosinophils 1.4%, monocytes 5.8% and basophils 0.2%. Erythrocyte Sedimentation Rate (ESR) was not elevated - 15mm/hr - and Mantoux test showed a negative response. Human Immunodeficiency Virus 1 and 2 testing was done and found to be negative. Sputum culture tested negative for Acid-Fast Bacilli.

***Corresponding author:**
Dr. Prasanna Kumar Saravanam
Associate Professor, Department of ENT, Head & Neck Surgery, Sri Ramachandra Medical College and Research Institute, Porur, Chennai-600116, India

Further evaluation with Contrast enhanced Magnetic Resonance Imaging (MRI) of the head and neck showed a "well circumscribed bilobed cystic lesion in the right masseter muscle extending to the right zygomatic region with post contrast peripheral rim enhancement." A probable diagnosis of "atypical complicated cyst suggestive of Hydatid Cysticercosis" was suspected by the Radiologist following ultrasonographic correlation. Immunological serology for Echinococcus was done and found negative.

Chest X ray PA view showed minimal Left Pleural Effusion. Fine Needle Aspiration Cytology (FNAC) of the swelling revealed "sheets of inflammatory cells composed of polymorphs and epithelioid macrophages in a necrotic background" suggestive of a granulomatous lesion. FNAC smear stained positive for Acid Fast Bacilli. Aspirated pus following FNA showed no growth in culture.

A diagnosis of cold abscess of the cheek was made and the patient was started on anti-tubercular chemotherapy as per the guidelines of the Revised National Tuberculosis Control Program – Isoniazid (600mg), Rifampicin (450mg) and Pyrazinamide (1500mg) three doses per week for first two months; followed by Isoniazid (600mg) and Rifampicin (450mg), three doses per week for next four months. Follow up showed complete regression of the swelling after one month of anti-tubercular chemotherapy.

Figure 1: Black arrow showing - 4cm x 2cm bilobed swelling over the right zygomatic arch

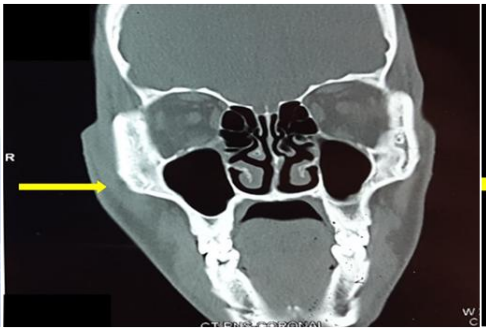

Figure 2: CT PNS – coronal section showing extent of lesion

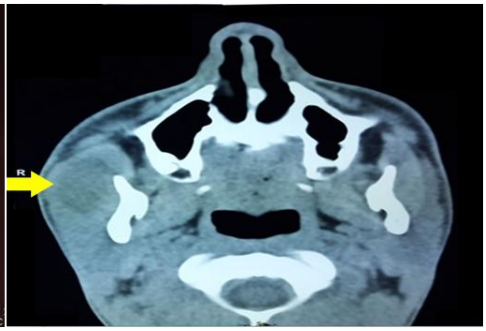

Figure 3: CT PNS – axial section showing a hypodense lesion over the right masseter region

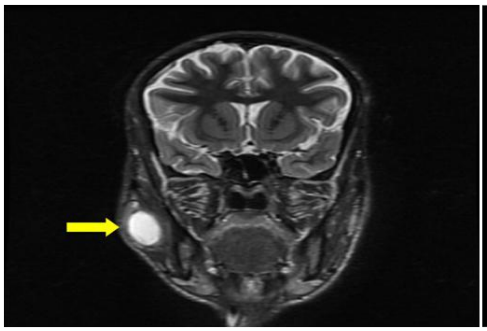

Figure 4: MRI head and neck coronal section

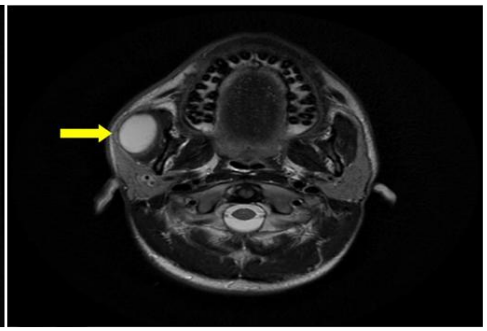

Figure 5: MRI head and neck axial section

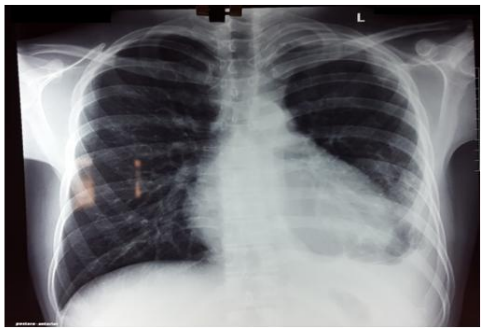

Figure 6: Chest X-ray PA view showing fluid collection with blunting of left costophrenic angle indicative of Left Pleural Effusion

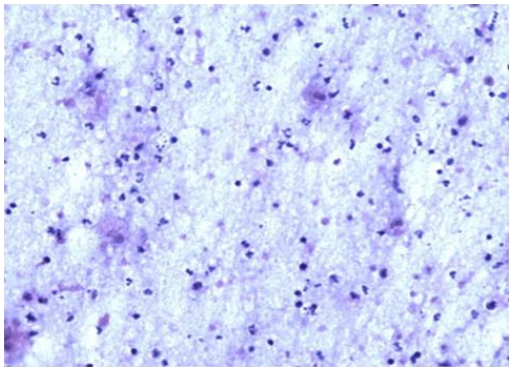

Figure 7: High power photomicrograph showing sheet of inflammatory cells (haematoxylin and eosin, x200 magnification)

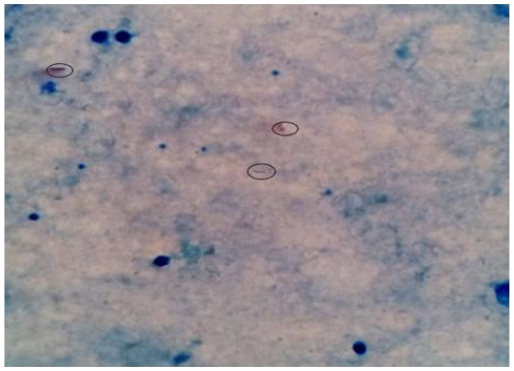

Figure 8: High power photomicrograph showing acid fast bacilli stained using Ziehl-Neelsen technique

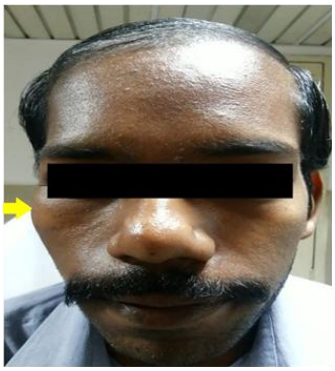

Figure 9: Complete regression of swelling as compared to pre-treatment photo following one month of anti-tubercular chemotherapy

DISCUSSION

The development and successful widespread implementation of Bacille Calmette-Guerin (BCG) vaccination coupled with an age of improved public health care and awareness, has aided in lowering the incidence of Pulmonary Tuberculosis. However, a concurrent pandemic of Human Immunodeficiency Virus (HIV) infection, and the widespread occurrence of Acquired Immunodeficiency Disease Syndrome (AIDS) has caused a hindering rise in Extrapulmonary Tuberculosis. Co-infection with HIV increases the risk of haematogenous dissemination

of the Mycobacteria and the resultant association between the two. Our patient was HIV negative, apparently immunocompetent individual, with an initial presentation of a cystic bilobed swelling over the right cheek. 60% of patients with extra-pulmonary disease are usually AFB sputum negative or have no chest radiographic evidence of pulmonary disease. It can present as a part of primary or late infection and can also serve as a reactivation site. The most common sites for extra-pulmonary disease includes – lymphatics (40%), pleural (20%), and bones and joints (10%) [8].

Tuberculosis of the cheek is a rare entity by itself, and when it does occur, it is usually a secondary lesion with a primary focus elsewhere in the body. Majority of secondary tuberculous lesions occur due to reactivation of the latent primary infection, and the rest due to reinfection. Usually, the primary lesion is a cutaneous tuberculous lesion like Scrofuloderma, that spreads the infective pathogen to other extra-pulmonary sites like parotid, eye, brain, masseter [9] etc. In our case, we found a suspected primary focus in the pleura with the Chest Radiograph revealing minimal Left Sided Pleural Effusion, with an absence of a cutaneous or oral focus.

Pleural tuberculosis is a form of primary tuberculosis, predominately seen in males and in an age group lesser than 45 years. It is believed that the presence of a small focus in the subpleural area, provides an access for the mycobacteria following its' rupture. This leads to a delayed hypersensitivity reaction between the mycobacteria and the resultant T-cells in the pleural fluid. The resultant inflammation causes decreased capillary permeability in a background of altered lymphatic drainage, leading to pleural effusion. These individuals ideally present with acute episodes of non-productive cough, dyspnoea, fever, and pleuritic chest pain. Our case had no such typical symptoms at presentation, except for an obvious swelling of the right cheek and an incidental finding of Left Sided Pleural effusion, with a negative AFB sputum culture. Further imaging was done with aid of CT -PNS and contrast enhanced MRI scan of the head and neck region. Due to the fact that the mass showed characteristics of an atypical complicated cyst in the right masseter region on MRI, our initial differential involved Hydatid Cyst of the cheek. Serological testing for Echinococcus antibody detection in the form of ELISA was done and found negative. ELISA for Echinococcus Antibody detection has been found to have a high sensitivity [10], hence making the probable diagnosis of Hydatid Cyst of the cheek less likely.

Prompt and timely diagnosis of EPTB does not follow ideal logistics that can be relied upon, as in the case of pulmonary tuberculosis. Diagnosis relies highly on early suspicion, elimination of other differentials and highly dependent on available resources. Fine Needle Aspiration Cytology (FNAC) is one such resource that has been found both sensitive and specific, especially when coupled with Ziehl-Neelsen Staining, obviating the need for an excisional biopsy in order to obtain an accurate diagnosis [11]. It was Fine Needle Aspiration Cytology (FNAC) that provided us with a confirmative diagnosis of Tuberculous Granuloma, showing clusters of bacilli positive for acid fast smear in a background of necrotic granuloma. The patient was immediately started on anti-tubercular chemotherapy as per the guidelines of RNTCP (Revised National Tuberculosis Protocol) [12] for a period of 6 months with regular follow-up.

This is a rare case of a secondary extra-pulmonary lesion of tuberculosis that was managed by us in an individual who was not only HIV negative, but also did not present with the classical symptoms suggestive of tuberculosis. Our experience re-iterated the need for high index of clinical suspicion with aid of advanced imaging that helps in characterising the lesion and FNAC providing an accurate diagnosis.

CONCLUSION

In conclusion, tuberculosis of the head and neck should be treated with high vigilance and promptness despite its rare occurrence. With the rising trend of Extra-pulmonary Tuberculosis, individuals with atypical symptoms should be dealt with a high clinical suspicion and FNAC to aid in its diagnosis, despite a negative sputum culture for AFB. It should be kept in mind that extra-oral tuberculosis need not always present with a concomitant oral lesion. It is imperative for Otorhinolaryngologists to be aware of the varied presentation of tuberculosis in the head and neck region however rare the occurrence maybe.

Conflict of interest – All authors have none to declare.

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