International Journal of Advances in Medicine
Bajpai J et al. Int J Adv Med. 2018 Apr;5(2):457-459
http://www.ijmedicine.com
pISSN 2349-3925 | eISSN 2349-3933
DOI: http://dx.doi.org/10.18203/2349-3933.ijam20181090

Case Report

Primary tubercular submandibular abscess: a rare presentation in two years old girl

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Received: 01 January 2018
Accepted: 03 February 2018

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ABSTRACT

Extra pulmonary tuberculosis (TB) continues to be a serious problem in developing countries. The prevalence of extra pulmonary tuberculosis (EPTB) is higher in immunocompromised, especially human immunodeficiency virus (HIV) co-infected patients. The most common site of extra pulmonary tuberculosis is lymph node followed by pleura. Lymph nodes are pivotal component of immune system and hence they are affected in various conditions like infections, autoimmune disorder, malignancy However, tubercular affliction of submandibular lymph node is not common. Only a few cases of tubercular submandibular abscess in immune-competent children have been reported in literature. Here we report a case of two-year old girl with non-healing multiple submandibular lymph node abscess presenting as a primary tuberculosis that proved diagnostically challenging. She responded favourably to a five drug anti-tubercular regimen.

Keywords: Immunocompetent, Line probe assay, Mycobacterium tuberculosis, Primary tuberculosis, Submandibular lymphadenopathy

INTRODUCTION

Tuberculosis (TB) is a global health problem with about 9.6 million people suffering from tuberculosis worldwide and nearly 2.8 million in India itself.[1]

Forty percent population of our country is infected with tubercular bacilli and lung is the most common site for tubercular infection.[2] The most common manifestation of extra pulmonary tuberculosis is lymphadenopathy.[3] Extra pulmonary tuberculosis seen in 10 to 15% cases of tuberculosis and tubercular lymphadenitis is seen in nearly 35% of extra pulmonary TB.[4] Tuberculosis is most important cause of peripheral lymphadenopathy in developing countries, contributing up to 43% cases.[5] In HIV-positive patients, extra pulmonary TB may account for up to 53-62% cases of TB.[6] Extra pulmonary tuberculosis may exist with or without pulmonary involvement. The incidence of lymphadenitis has increased with the rising incidence mycobacterium tubercular infection worldwide.[7]

Cervical lymph node involvement seen in 60-90% of patient with or without other lymph node involvement. Tubercular lymphadenitis can be local manifestation of the systemic disease.[8]

It may occur during primary tubercular infection or as a result of reactivation of dormant foci or direct extension from a contiguous focus. Primary infection occurs on initial exposure to tubercle bacilli.[9] Even primary tubercular involvement is prevalent in youngsters and adolescents than in elderly.[10]

CASE REPORT

A two-year old girl from north India presented with complaints of left submandibular swelling for past three months. It was progressively enlarging in size and was associated with low grade fever and mild local pain. She had received multiple courses of antibiotics, without any meaningful improvement. There was no history of tuberculosis in her family.

On local examination, she had a non-tender, circumscribed, solitary swelling of dimensions - 6mm x 4mm in the left submandibular region. The swelling was firm in consistency with ill-defined borders. The skin over the swelling was intact and the temperature was not raised. A BCG vaccine scar was also noticed. Fine needle aspiration cytology and excision biopsy from the lesion was done but it was inconclusive. Despite multiple antibiotics regimens the wound was not healing, and pus discharge continued. After fifteen days her parents noticed another swelling in submandibular region. It was also soft with dimensions of 2 cm X 3 cm. Lesion was also extending towards cervical region. The rest of the head and neck examination was normal. Hematological and biochemical profile was normal. Chest X-ray did not show any abnormality (Figure 1). Fine needle aspiration cytology (FNAC) from the most prominent part of the swelling yielded purulent material. A smear stained with hematoxyline and eosin showed caseous necrosis and degenerated mixed inflammatory cells and few clusters of epitheloid cells, suggesting tuberculoid or caeseating granuloma (Figure 2). A Gram stain did not show any organism. A smear stained by Ziehl Neelsen technique showed acid-fast bacilli (AFB).

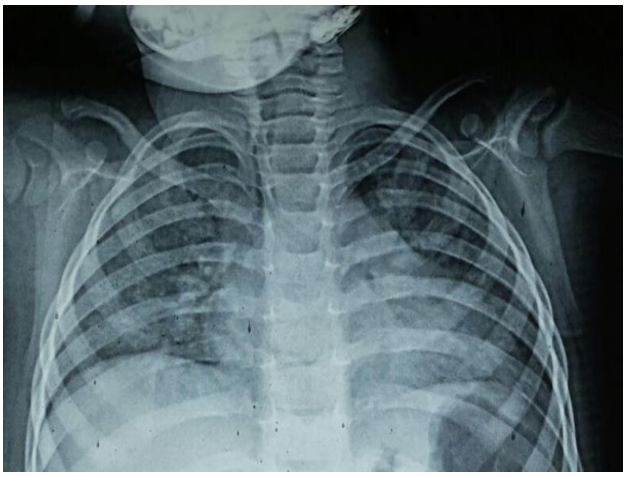

Figure 1: Chest X-ray AP view of two years old girl with no pleuroparenchymal disease.

The patient was treated with World Health Organization (WHO) approved directly observed treatment short course (DOTS) category I regimen under the Revised National Tuberculosis Programme. Despite four months of anti-tubercular therapy she was not responding to treatment, wound was still not healing and new lesions were also present (Figure 3). Again, pus was aspirated and sent for molecular method of testing- Line probe assay (LPA) and BACTEC culture which was positive for Mycobacterium tuberculosis (MTB) complex and sensitive to rifampicin and isoniazid. On the basis of this report and clinical presentation of patient, author made a diagnosis of treatment failure and started DOTS category II treatment. The swelling and fever had subsided after 3 months of category II treatment. Her appetite was normal, and she had gained 3kg in weight. There was no clinical evidence of disease at the end of nine months of Category II regimen.

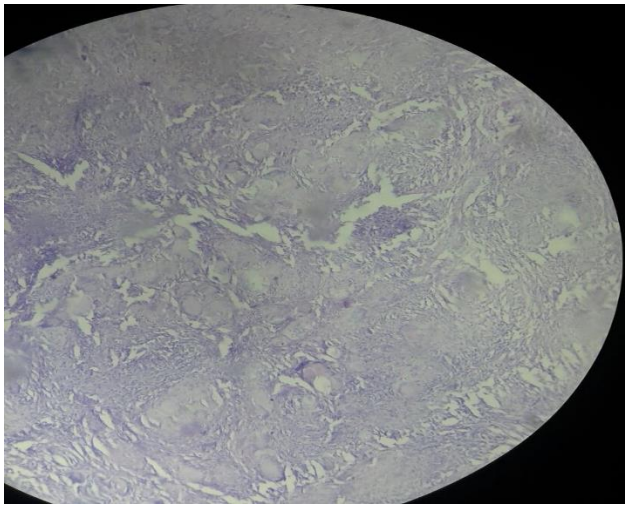

Figure 2: Fine needle aspiration cytology (FNAC) from the most prominent part of the swelling showed caseous necrosis and few clusters of epitheloid cells, suggesting tuberculoid or caeseating granuloma.

Figure 3: Left submandibular non-healing tubercular abscess after excision.

DISCUSSION

Primary tuberculosis usually manifests in lung, gastrointestinal tract and head-neck regions. Tuberculosis of the head and neck region comprises about 10% of all

the cases of extra-pulmonary tuberculosis. The cervical lymph nodes are the most commonly affected followed by laryngeal tuberculosis, deep neck space abscess and tuberculous otitis media.[11,12]

The diagnosis is difficult on clinical grounds because the swelling may be representative of benign, malignant and tubercular, non-tubercular mycobacteria (NTM) or bacterial infections. Reports of primary submandibular TB in a two-year old girl are sparse.[13] NTM are also causative organism in extrapulmonary tubercular cases.[14] In the present case a swelling was present in left submandibular area and on oral examination no obvious lesion was present which could be the cause of the swelling. A panoramic radiograph of the affected area was taken to check for any underlying source of intraoral involvement with respect to the swelling; it did not reveal any odontogenic origin in relation to the swelling and thus it was determined that the swelling was nonodontogenic in origin.[15] The presence of matting in the mass of the swelling and non-odontogenic nature of the swelling was taken into consideration for a clinical diagnosis of right submandibular tuberculous lymphadenitis. Molecular methods like LPA and CBNAAT allow for rapid diagnosis Tuberculosis in the cases of extrapulmonary involvement. In addition, they allow for drug sensitivity testing too. In our case, LPA of pus aspirated after 4 months of Cat I DOTS therapy was positive. Hence, author switched to Cat II DOTS regimen with complete clinical response.

The diagnosis is usually made either by a process of excision or following histopathology of the surgically extirpated specimen. A high index of suspicion, rapid diagnosis, treatment with anti-tubercular drugs, and minimal intervention can successfully manage the disease without the risk of major surgery and thereby maintain the cosmesis. Advanced microbiological tools can give the additional benefit of rapid diagnosis and culture specific drugs for targeted therapy.

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

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Cite this article as: Bajpai J, Kant S, Verma AK, Bajaj DK, Pradhan A. Primary tubercular submandibular abscess: a rare presentation in 2 years old girl. Int J Adv Med 2018;5:457-9.