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Case Report

Tuberculous Lymphadenitis in Submandibular Area: A Case Report

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Received: January 24, 2023; **Published:** January 31, 2023

Abstract

Tuberculous lymphadenopathy is a common cause of pediatric peripheral lymphadenopathy.

Lymph node involvement is the common clinical symptom of extrapulmonary tuberculosis in the pediatric patients. We present a 2-year-old female patient with previous history of diffuse hard painless right submandibular swelling with purulent discharge. She underwent previous surgical drainage. The patient was diagnosed with right submandibular tuberculous lymphadenitis based on previous histopathological report. She came for following with us at pediatric surgical outpatient clinic with clinical symptom of nonhealing wound with purulent discharge after surgical drainage at right submandibular region.

Keywords: *Submandibular Tuberculous Lymphadenitis; Tuberculosis; Tubercular Lymphadenitis; Giant Cells; Mycobacterium tuberculosis*

Introduction

Tuberculosis is the most common systemic bacterial infectious diseases [1]. It is the cause of persistent cervical lymphadenopathy [2]. It was defined as a painless soft swelling in a group of superficial regional lymph nodes caused by *Mycobacterium tuberculosis* [3,4]. Extrapulmonary tuberculosis is rare with incidence 0.05 - 5% of tuberculosis patients [4]. We report the case with aim to focus to this pathology by presenting a case of a pediatric patient diagnosed with submandibular tuberculous lymphadenitis.

Case Report

A 2-year-old female patient presented with painless swelling within her right submandibular region (Figure 1). She underwent the first surgical drainage in this region at the hospital A. She was diagnosed with right submandibular tuberculous lymphadenitis based on previous histopathology report. The first-line antituberculous therapeutic drug was administered for 4 months. At post drainage 1 month, the progression of postsurgical drainage wound at right submandibular region was not improved. It remained purulent discharge from previous surgical drainage site and there was nonintact skin. Her guardian brought her to our hospital with a problem of nonhealing wound after surgical drainage at right submandibular region. On examination, there was a slightly nontender firm right-sided submandibular swelling nearly 5 cm. The superimposing skin was intact with erythematous color compared to the surrounding skin. Palpable pathologically enlarged right submandibular regional lymph nodes including bilateral cervical lymph nodes were evident. There was no clinical symptom in dysphagia nor odynodysphagia. The patient denied recent fever, upper respiratory infection, B-symptoms or constitu-

tional symptoms. She denied history of recent sick contact, travelling and or any animal contacts. Her chest X-ray was performed and the result was in a limit of normal impression. Ultrasound examination was performed focusing on cervical lymph nodes exhibited the size of the expanded lymph nodes with increased hypervascularity. Within 24 hours following the hospitalization the swelling got fluctuant and the patient was post for incisional and drainage under general anesthesia. Excision specimen was submitted for histopathological and microbiological examination. The histopathological examination of the lymph node biopsied example showed an evidence of ill-defined infiltrated by coalescent epithelioid histiocytic granuloma with areas of central caseous necrosis (Figure 2). The report revealed tuberculous lymphadenitis. On additional history taking, patient's grandfather gave a history of pulmonary tuberculosis when the patient was in previous exposure with him at the age of 1 year old. So, a clinical diagnosis of a right submandibular tuberculous lymphadenitis was ponders and confirmed by positive real time polymerase chain reaction (PCR) for *Mycobacterium tuberculosis* complex (MTBC). The wound at operative field healed uneventful. The patient was referred to the pediatric department for following clinical symptom with continuing antituberculous therapeutic regimen.

Figure 1: *A diffuse swelling in right submandibular region.*

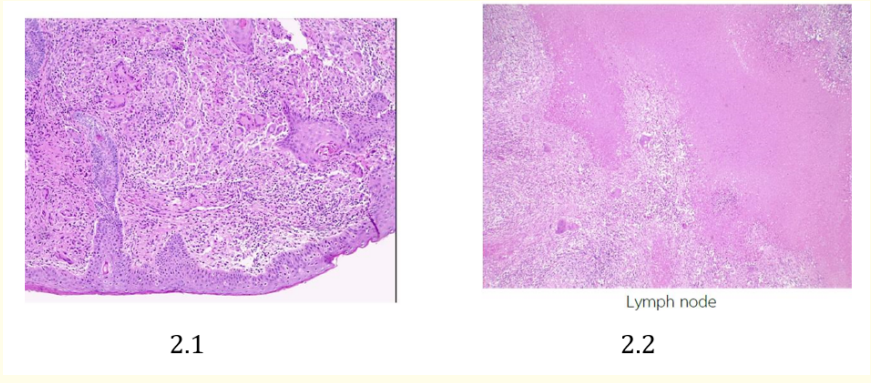

Figure 2: *Low power view showing a granuloma composed of epithelioid histiocytes with abundant eosinophilic cytoplasm and multinucleated giant cell formation, consistent with a tuberculous etiology (hematoxylin-eosin stain, X 40).*

Figure 2.1: *Histology of skin [H&E section, magnification 10X10] showing granulomatous dermatitis.*

Figure 2.2: *Histology of lymph node [H& E section, magnification 10X10] showing granulomatous lymph node.*

Discussion

Tuberculosis is a chronic granulomatous disease caused by different strains of mycobacteria, especially *Mycobacterium tuberculosis* in human [5]. The diagnosis of extrapulmonary tuberculosis is often overlooked because it has no specific pathognomonic signs [6]. A new classification system of orofacial tuberculosis in different forms and locations is presented [6] (Table 1). The most common form of extrapulmonary tuberculosis is cervical tuberculous lymphadenitis or scrofula [7]. It had female predilection in children and young adults [8], as in our case. According to the proposed five stages classification involving the progression of tuberculous lymphadenitis which has been described by Jones and Campbell [9] (Table 2), our patient can be classified as type IV stage 3, where there was abscess formation.

Type I	Lumpy jaw: Patient presents with extraoral swelling without any intraoral or extraoral draining sinuses; the focus of infection involves the mandible or maxilla; in general, the patient's oral hygiene is good.
Type II	Patients report a history of extraction and present with nonhealing extraction sockets with/ without intraoral or extraoral draining sinus/ sinuses.
Type III	Patients report no history of extraction and present with intraoral or extraoral draining sinus/ sinuses in the orofacial region and an osteomyelitic bony lesion.
Type IV	Tuberculous lymphadenitis of the head face neck region without any features of type I, II, III, or V.
Type V	Lesion of other sites in and around the oral cavity, eg, maxillary antrum, salivary glands, orofacial muscles, gingiva, tongue, etc.

Table 1: *Andrade's classification for orofacial tuberculosis [6].*

Stage 1	Enlarged, firm, mobile, discrete nodes showing non-specific reactive hyperplasia
Stage 2	Large rubbery nodes fixed to surrounding tissue owing to periadenitis
Stage 3	Central softening due to abscess formation
Stage 4	Collar-stud abscess formation
Stage 5	Sinus tract formation

Table 2: *Stages of progression of tuberculous lymphadenitis - Jones and Campbell classification [9].*

The diagnosis of tuberculous lymphadenitis needs a histological examination of tissue and demonstration of the infective organisms in the specimen [8]. The histological picture of tuberculosis is characterized by presence of both caseous and non-caseous granulomas with multinucleated giant cells [8]. In our case, the diagnosis of tuberculosis was suggested by caseous granuloma on histology from the biopsy result and confirmed with a positive polymerase chain reaction (PCR). The Infectious Disease Society of America (IDSA) guidelines recommend surgical excision only in unusual condition [10]. Paradoxical upgrading reaction (PUR) is a frequency which patients experience clinical symptoms of worsening during treatment of tuberculous lymphadenitis [11]. Manifestation of PUR in our patient included enlarging lymph nodes and had discomfort from tense, fluctuation of the lymph nodes resulting in incision and drainage with surgical excision to be determined and performed in our patient in order to short duration period of PUR [12].

Conclusion

Identification of tuberculosis especially isolated extrapulmonary manifestation can become challenging. Atypical presentations would make clinician especially in developing countries should be always kept in mind and make identification of pathology toward the correct diagnosis and curative treatment.

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Volume 12 Issue 2 February 2023