CASE REPORT

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A rare association of tonsillar tuberculosis and lichen scrofulosorum



Anshika Harit^{1*}, Anjan Kumar Sahoo² and Ishwar Singh²

Abstract

Background: Both tonsillar tuberculosis and lichen scrofulosorum are sporadic presentations of extrapulmonary tuberculosis. Lichen scrofulosorum commonly presents in children and young adults as lichenoid eruptions over the skin. Granulomatous inflammation of the tonsils, however, presents as non-specific sore throat and foreign body sensation in the throat. The concomitant presentation of the abovementioned tubercular manifestations has not been reported in the literature.

Case presentation: Herein, we report a case of an 11-year-old male patient who presented with a history of recurrent sore throat and ulcerative lesion over the tonsil. Systemic examination revealed multiple perifollicular eruptions over the trunk and back. The diagnosis was confirmed on histopathological findings of epithelioid cell granulomas with Langerhans giant cells following biopsy from the tonsil and skin lesions. Antitubercular therapy was initiated soon after. The patient responded to treatment as early as 6 weeks and was completely asymptomatic at 1 year of follow-up.

Conclusion: A diagnosis of granulomatous tonsillitis should alert the physician to the possibility of underlying systemic tuberculosis. In our case, coexistence of lichen scrofulosorum helped us to substantiate the diagnosis based on the biopsy report. Response to antitubercular agents is excellent and should be started at the earliest.

Keywords: Tuberculosis, Cutaneous, Palatine tonsil, Antitubercular agents, Diagnosis, Differential

Background

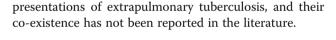
Granulomatous inflammation of the tonsils is rare, and its presence should alert the clinician to the possibility of a subjacent systemic disease [1]. Tuberculids are cutaneous immunological manifestations of pulmonary or extrapulmonary tuberculosis. Lichen scrofulosorum (LS) is one such rare form of tuberculid, characterized by lichenoid eruptions over the skin. The cutaneous lesions are majorly distributed around the hair follicles and may be scattered or grouped over the trunk, back, and extremities. LS can be considered as a crucial marker of occult tuberculosis. Immunocompetent children with underlying TB are frequently affected [2]. Both tuberculosis of tonsil and lichen scrofulosorum are sporadic

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Case presentation

An 11-year- old male presented to the ENT OPD with the chief complaint of tightening of the skin over the face and nape of the neck for the past 8 months (Fig. 1), with an associated complaint of swelling over the neck for 15 days. The patient also complained of recurrent sore throat and fever, which gradually subsided. The family history of TB was positive. On examination, bilateral grade 4 tonsillar hypertrophy was seen (Fig. 2) and an ulcerative lesion over the left tonsil. The patient had a problem with mouth opening due to the stretched skin over the face. Multiple discrete lymph nodes were also palpated in the submental region (Fig. 3). FNAC from submental swelling was suggestive of non-specific inflammation.

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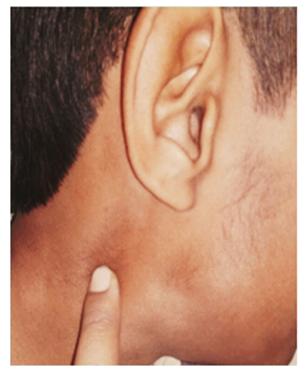


Fig. 1 Stretched skin



Fig. 2 B/L grade IV tonsillar hypertrophy

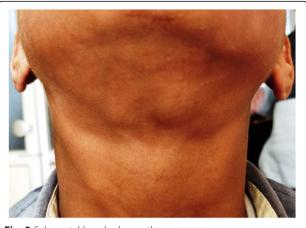


Fig. 3 Submental lymphadenopathy

The patient was planned for a tonsillar biopsy which showed multiple well-defined epithelioid cell granuloma admixed with multiple giant cells suggestive of granulomatous inflammation. Routine blood investigations were within normal limits except for ESR (36 mm in 1st hour). Biochemical tests like CRP, c-ANCA, p-ANCA, and rheumatoid factor to rule out autoimmune conditions were negative, but the Montoux test was reactive $(23 \times 15 \text{ mm})$ at 48 h (Fig. 4). On anamnesis, the patient revealed he had lesions over his chest and back for the same duration, which were painless. On physical examination, dome-shaped skin-colored lesions in follicular and parafollicular distribution were seen over the trunk and back (Figs. 5 and 6). LS was kept as the first



Fig. 4 Reactive Montoux, 23 × 15 mm



Fig. 5 Multiple discrete as well as grouped skin-colored lesions over the trunk and back



Fig. 6 Multiple discrete as well as grouped skin-colored lesions over the trunk and back

differential, and a skin biopsy was carried out to confirm the suspicion. Biopsy showed epithelioid cell granulomas with Langerhans giant cells in the papillary dermis; Ziehl-Neelsen (ZN) stain for AFB was negative. Biopsy findings were consistent with the diagnosis of lichen scrofulosorum.

The patient was started on antitubercular therapy. A 6-month regimen with first-line drugs consisting of rifampicin (10 mg/kg/day), isoniazid (5 mg/kg/day), ethambutol (25 mg/kg/day), and pyrazinamide (30 mg/kg/day) for 2 months followed by isoniazid and rifampicin for 4 months (2RHZ + 4RH) was given. The lymph nodes regressed, and the skin lesions resolved in 6 weeks. The patient was followed up for 1 year and was found asymptomatic at the last follow-up.

Discussion

The presence of granulomatous tonsillitis raises the possibility of an underlying concurrent clinical illness. It is most commonly seen in TB, followed by sarcoidosis, fungal infection, Hodgkin's disease, keratinizing squamous cell carcinoma, and tonsillar malakoplakia [3]. Sore throat, dysphagia, and cervical lymphadenopathy are typical features in a patient with tonsillar tuberculosis [4]. It could resemble sarcoidosis and thus can be difficult to diagnose if the culture for AFB is negative. Tonsillar biopsy in such cases will reveal granulomatous inflammation characterized by caseating epithelioid granulomas with Langhans' and foreign body giant cells. On the other hand, non-caseating granulomas are a feature of sarcoidosis [5].

Tuberculids are a result of a delayed cutaneous immunological reaction to tubercular bacilli [6]. Lichen scrofulosorum is a form of tuberculid. Papular lesions of LS display a strongly positive tuberculin reaction and are most commonly exhibited by children and young adults. The patient presents with minute papules on the back, trunk, and extremities. They are painless, skin-colored to hyperpigmented raised lesions that may be scattered or grouped with para-follicular distribution [2]. Acid-fast bacilli are not detected in LS lesions as they are probably destroyed due to the hypersensitivity reaction [7]. Differential diagnoses should exclude other causes of dermatoses such as keratosis pilaris, lichen spinuloses, lichen nitidus, pityriasis rubra pilaris, and lichenoid sarcoidosis. Although LS is a rare occurrence, it is an important marker of occult tuberculosis. Standard treatment is to start ATT for the patient with four drugs [8]. Our patient's initial histopathology report revealed non-specific granulomatous tonsillitis, with negative ZN stain and culture for AFB. However, the diagnosis of LS was later confirmed on skin biopsy. The absence of pulmonary involvement was an unusual feature in our case and pointed us towards our diagnosis. The association of tonsillar hypertrophy with ulceration and LS is intriguing. Nevertheless, an unstable immunological state might have influenced the unusual expression in our patient.

Conclusion

The clinician needs to consider all the possible diagnoses when dealing with unusual symptoms, and a thorough systemic examination is a must as not to miss out on rare presentations.

Both tuberculosis of tonsil and lichen scrofulosorum are infrequent scenarios of extrapulmonary tuberculosis. These differentials should be kept in mind in patients who present with typical symptoms like sore throat, recurrent tonsillitis, and skin lesions. A broader view while tackling these patients and a multidisciplinary approach will help in reaching a proper diagnosis.

Abbreviations

ATT: Antitubercular treatment; c-ANCA: Cytoplasmic-antineutrophil cytoplasmic antibodies; CRP: C-reactive protein; ESR: Erythrocyte sedimentation rate; FNAC: Fine-needle aspiration cytology; LS: Lichen scrofulosorum; p-ANCA: Perinuclear-antineutrophil cytoplasmic antibodies; TB: Tuberculosis; ZN: Ziehl-Neelsen

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Authors' contributions

AH was responsible for gathering the data and writing the manuscript. AKS revised the draft and contributed in preparation of the final version. IS approved the final version of the manuscript. All authors have read and approved the final manuscript.

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Availability of data and materials

Patient's personal details have not been made available due to privacy concerns.

Ethics approval and consent to participate

The Otolaryngology Department board of Maulana Azad Medical College & Lok Nayak Hospital, Delhi, India approved the case study. Consent to participate is not applicable.

Consent for publication

Informed written consent was obtained from the patient's father for publication of this case report and any accompanying images. The copy of the signed consent form is available should the journal wish to review it.

Competing interests

The authors declare that they have no competing interests.

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