

Case Report

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Atypical tonsillitis in a teenager: A case report**J Oubenhah***; B Hemmaoui; B Amrani; A Ftouhi; F Benariba; N Errami

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Abstract

Chronic tonsillitis may be caused by different pathogens. One of them is tubercle bacilli. Tuberculosis is one of the major infectious diseases worldwide. Although, tonsillar tuberculosis is very rare manifestation of extra-pulmonary tuberculosis, and it is classified into primary tuberculosis of tonsils, occurring in the absence of active pulmonary tuberculosis, and secondary tuberculosis of tonsils in which an associated pulmonary tuberculosis is documented. We report the case of 13 years-old female teenager with no medical history, presenting chronic tonsillitis with 3-4 months' complaint of sore throat, dysphagia, odynophagia, low-grade fever and weight loss, for whom the examination of oral cavity revealed bilateral asymmetrical tonsils enlargement, more marked on the right side, associated with ulcerations, simulating malignancy. She received several antibiotics during this period, with no improvement. Bilateral tonsillectomy was performed and typical features of tonsillar tuberculosis was revealed by histopathological examination. No pulmonary tuberculosis was documented and the patient was HIV seronegative. The diagnosis of isolated and primary tonsillar tuberculosis was established in an immunocompetent pediatric patient. She was successfully treated with anti-tuberculosis therapy for 6 months.

Keywords: Chronic tonsillitis; Tonsillar tuberculosis; Extrapulmonary tuberculosis; Histopathology; Pediatrics.

Introduction

Tonsillitis is very common disease among school-aged children, yet it can manifest in individuals of any age [1]. Tonsillitis refers to the inflammation of the palate tonsils [2], which are situated within the Waldeyer lymphatic ring [1]. The clinical presentation of acute tonsillitis is a variable combination of symptoms, including a sore throat, difficulty and pain during swallowing, fever, chills, asthenia, and reflex otalgia. Additionally, nausea, vomiting, abdominal pain, halitosis, and mouth opening limitation may also be present [3]. Physical examination usually shows enlarged and erythematous tonsil with the presence of visible purulent material or cheesy exudate on the tonsillar surface, accompanied by cervical lymphadenopathy [3]. The term of "recurrent acute tonsillitis" is used to designate

the occurrence of repeated episodes of sore throat interrupted by intervals with no or insignificant symptoms [4]. Chronic tonsillitis, on the other hand, is defined as a sore throat that has been evolving for at least 3 months, associated with clinical signs of tonsil inflammation, that improve following antibiotic treatment but without complete resolution [5]. Viral pathogens such as adenovirus, rhinovirus, enterovirus, influenza virus and Epstein-Barr virus are involved in 70-95% of tonsillitis cases [6]. Bacterial agents are responsible of 15-30% of cases. The most common bacterial pathogen is Group A β -hemolytic Streptococcus pyogenes [6]. Other bacteria may also be responsible, such as Staphylococcus aureus, Haemophilus influenzae, Streptococcus pneumoniae, Escherichia coli [7]. In less common cases, other bacteria may be involved, including Corynebacterium diphtheria, and sexually transmitted bacteria such as Neisseria

gonorrhoeae and *Chlamydia trachomatis* [1]. In extremely rare cases, tonsillitis is a form of extrapulmonary tuberculosis [8].

We report a case of primary tonsillar tuberculosis in a 13 years-old immunocompetent female teenager, in order to provide insights into the clinical presentation of this rare extrapulmonary manifestation of tuberculosis and to emphasize the need for evoking this diagnosis in the presence of chronic tonsillitis, especially in countries where tuberculosis is still endemic.

Case presentation

A 13-year-old female teenager was referred to our department with the complaint of sore throat, dysphagia, odynophagia, and low-grade fever for the last 3-4 months. The onset was insidious, and evolution was gradually progressive. Her parents reported the presence of weight loss of approximately 3 kg, bad breath, snoring, and mouth breathing. There was no history of cold, cough or any other chest symptoms. She received several antibiotics (Amoxicillin, Amoxicillin plus clavulanic acid, Azithromycin, Cefixime) during this period, with no improvement. The patient has been vaccinated according to the national vaccination program, which includes the *Bacillus Calmette-Guérin* (BCG) vaccine for tuberculosis, and has no previous medical history. Past or family history of tuberculosis was not present. On general examination, the patient seemed of a healthy form. Oral examination revealed bilateral tonsillar grade IV asymmetrical enlargement, more marked on the right side, associated with ulcerations, cheesy pus and erythema of anterior pillars (Figure 1). The posterior wall of the oropharynx was of normal aspect. The examination of the cervical region revealed the presence bilateral cervical lymphadenopathy at levels II and III, which were enlarged, tender, and of firm consistency. Detailed examination has not revealed any other abnormal findings. Nasal cavity, nasopharynx, base tongue and larynx were clear on the complete nasofibroscope.

On routine laboratory investigations, Hemoglobin was 13.2 g/dl, Total leucocyte count was 9,700 cells/mm³, and C reactive protein was 11 mg/l. Liver and renal function tests were within normal ranges. Her chest X-ray was normal. Ziehl Neelsen and Gram staining of sputum and throat swab were negative as well as pyogenic cultures.

In order to establish a definite diagnosis and exclude malignancy, a bilateral tonsillectomy was performed, and the specimen was sent to pathology. Histopathological examination revealed on both tonsils, the presence of multiple inflammatory granulomas with epithelioid cells, Langhans giant cells and caseous necrosis, associated with acid fast bacilli on Ziehl Neelsen staining. These features were compatible with the diagnosis of tonsillar tuberculosis. Immunohistochemistry was done to rule out the possibility of association with malignancy and showed negative reaction for anti-Ki67 and anti-p53. Acquired immunodeficiency syndrome was excluded by negative HIV serology test. The definite diagnosis for this case was primary tonsillar tuberculosis in a immunocompetent young patient. She therefore received an anti-tuberculosis treatment protocol consisting of 2 months of daily Rifampicin (10 mg/kg), Isoniazid (5 mg/kg), Ethambutol (15 mg/kg), and Pyrazinamide (25 mg/kg), followed by 4 months of Rifampicin (10 mg/kg) and Isoniazid (5 mg/kg). The patient's symptoms improved well at the 1-month follow-up, and the tonsillar beds were of normal aspect.



Figure 1: Bilateral asymmetrical tonsils enlargement, more marked on the right side, associated with ulcerations, cheesy pus and erythema of anterior pillars.

Discussion

Tuberculosis is an infectious disease caused by bacilli of the mycobacterium genus [9]. It remains a significant global health problem, as it is the most prevalent infection and it is ranked among the top ten causes of death due to infections in the world [10,11]. According to the World Health Organization's estimations published in the Global Tuberculosis Report 2023, 10.6 million people developed tuberculosis in 2022, 12% of whom are children under the age of 14, and 1.3 million people died from the infection [12]. Globally, the estimated incidence rate of tuberculosis is 133 per 100,000 population per year [12]. Morocco is listed among countries having an intermediate incidence of tuberculosis, with an estimated incidence rate for 2022 was 93 per 100,000 population per year [12]. Although tuberculosis primarily affects the lungs, 16% of cases are extra-pulmonary [13], causing approximately 25% of overall tuberculosis morbidity [14]. The lymph nodes are the most commonly affected site, followed by the pleura. Other sites are known to be affected, such as the central nervous system, the musculoskeletal system, the gastrointestinal tract, the genitourinary system, and the peritoneum the pericardium [13]. Tuberculosis of the oral cavity is rare, accounting for less than 0.5-1% of all tuberculosis cases [15]. The tongue is the principal affected site of involvement, nevertheless, any oral mucosal such as the palate, lips, buccal mucosa, gingiva, palatine tonsils and floor of the mouth may also be involved [13-15]. Tonsillar tuberculosis is therefore extremely rare, and this may be explained by local antibacterial properties, mainly due to the antiseptic and cleansing action of saliva, to the thickness of stratified squamous epithelial lining of the tonsil, and also to the presence of saprophytic organisms in the oral cavity which prevents from colonization by tubercle bacilli [16].

Tonsillar tuberculosis is classified into primary tuberculosis of tonsils, which occurs in the absence of active pulmonary tuberculosis, and secondary tuberculosis of tonsils in which an associated pulmonary tuberculosis is documented [8]. Primary tonsillar tuberculosis is attributed to the consumption of unpasteurized bovine milk contaminated with *Mycobacterium bovis*, whereas secondary tonsillar tuberculosis is mostly due to direct inoculation by *Mycobacterium tuberculosis* present in expectorated infected sputum or inhaled infected droplet nuclei [13-17]. It may also be caused by hematogenous spread from

a primary site [18]. It has been reported that isolated tonsillar tuberculosis is a rare occurrence typically seen in the younger populations, and secondary tuberculosis of tonsils is more common. Nevertheless, Moisa et al. reported in their literature review that there were more cases of primary tonsillar tuberculosis than secondary forms (59% vs 41%) [19], which is a surprising finding in our era, where pasteurization of milk is very widespread. In addition to known risk factors of tuberculosis, there are predisposing factors of tonsillar tuberculosis including poor oral hygiene, leukoplakia, local trauma, dental extraction, periodontitis and irritation by clove chewing [19,20]. However, none of these factors was present in our case. Earlier epidemiologic data stipulated that there is no age or sex predilection in cases of tonsillar tuberculosis [8-21]. Nevertheless, more recent data shows a predominance of male cases (67,25%), and the age of occurrence varied from 3 to 81 years old, with almost two-thirds of cases being 20 to 50 years old [19]. Tonsillar tuberculosis presents as chronic tonsillitis, the commonest symptoms of which are sore throat, dysphagia and odynophagia, accompanied or not by tuberculosis constitutive symptoms such as low-grade fever, weight loss, night sweats [13]. Physical examination of the oral cavity usually shows enlargement of one or both palatine tonsils, with whitish to yellow plaques and superficial or deep ulcerations on their surface [8-22]. In 46 % of cases, tonsillar tuberculosis is bilateral [19], with asymmetrical enlargement [8]. This last feature is reported to be suggestive of tuberculosis of tonsils, as well as tonsillar enlargement without exudate, painful deglutition, obliteration of crypts, and the presence cervical lymphadenopathy [16]. It is certainly difficult to evoke the diagnosis of tonsillar tuberculosis only on clinical features, particularly in the absence of other forms of tuberculosis. Traumatic ulceration, aphthous ulcerations, actinomycosis, syphilis, severe fungal disease, Plaut-Vincent's tonsillitis, Wegener's granulomatosis, lympho-reticular malignancy, and squamous cell carcinomas are the principal differential diagnosis [23]. The definite diagnosis of tonsillar tuberculosis is made by identifying *Mycobacterium tuberculosis* in the diseased tonsils, or by the histopathological examination of tissue samples obtained by punch biopsy of the lesion or after tonsillectomy [8-21]. In the review of 86 cases, Moisa et al. reported that the diagnosis was made in all patients on the basis of histopathology [19]. This may be explained by the rarity of this disease and the clinicians' need to rule out malignancies, especially with the persistence of symptoms, weight loss, and the presence of ulcerations, or in the case of tonsillar tuberculosis diagnosis is not evoked. Typical histopathological features of tonsillar tuberculosis include epithelioid cell granulomas with caseous necrosis, Langhans and foreign body giant cells with or without acid fast bacilli [17]. Gene analysis by PCR could be used to obtain confirmation of diagnosis [13-23]. In all cases, sputum smear and chest X-ray should be done to rule out pulmonary involvement, and the HIV status should also be screened [8]. The management of tonsillar tuberculosis is similar to other forms of tuberculosis. It consists on administration of multi-drugs antitubercular therapy which includes 4 antibiotics: Rifampicin (10 mg/kg), Isoniazid (5 mg/kg), Ethambutol (15 mg/kg), and Pyrazinamide (25 mg/kg), daily for 2 months, followed by Rifampicin (10 mg/kg), Isoniazid (5 mg/kg), daily for 4 months [24]. This treatment was successful for our patient, who was symptoms free on the follow-up. Our case patient underwent a bilateral tonsillectomy with histopathological examination because of the unusual presentation and in order to exclude the possibility of malignancy. That leads to the discovery of tonsillar tuberculosis. Although, we consider that tonsillectomy should be part of the manage-

ment alongside medical treatment in cases where the diagnosis is made without recourse to removal of the tonsils, because it will help to eradicate the tubercle bacilli.

Conclusion

Tonsillar tuberculosis is a rare entity, particularly in pediatric population. The diagnosis is challenging, but through this case, we think that any case of symptomatic chronic tonsillitis, especially when associated with weight loss, should raise suspicion of tonsillar malignancy in the first place, followed by tonsillar tuberculosis. Biopsy sample should be taken and fractionated. The first fragment is sent to histopathology, and the second one is placed in a dry tube and sent to bacteriology for staining, culture and gene PCR analysis. The treatment is based on anti-tuberculosis therapy. The treatment is frequently successful, with symptoms disappearing within a few weeks. Even with well-managed treatment, there remains a risk of relapse or failure to recover. These failures are caused by the emergence of anti-bacillary resistant BK strains, low serum concentration of anti-tuberculosis drugs and the non-compliance with medication.

Conflicts of interest: The authors declare that they have no conflict of interests.

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