INTERNATIONAL JOURNAL OF STUDENTS' RESEARCH
Volume 2 Issue 1 Year 2012 **www.ijsronline.com**

CASE REPORT

Unilateral tonsillar actinomycosis masquerading as a neoplasm

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ABSTRACT

Actinomycosis is an endogenous infection caused by gram positive bacteria with filamentous appearance, of the genus Actinomyces, of which the most common pathogen is Actinomyces israelii. Tonsillar actinomycosis is usually a bilateral feature. Although actinomycosis can be detected in the tonsils comparatively frequently, there are only few reports regarding unilateral tonsillar hypertrophy due to actinomycosis. To our knowledge, only six cases of unilateral tonsillar hypertrophy caused by actinomyces infection have been reported until now. Four of them were masquerading as a tumor, and two as pendulous masses attached to the tonsils. We herein report a case of unilateral actinomycosis masquerading as a tonsillar neoplasm in an 18 year old boy.

Key Words: Actinomycosis, tonsil, neoplasm

Introduction

Actinomycosis is an endogenous infection caused by gram positive bacteria with filamentous appearance, of the genus Actinomyces, of which the most common pathogen is Actinomyces israelii [1]. Although on culture, Actinomyces species were originally classified as fungi because of their branching structures, they are true bacteria with neither mitochondria nor nuclear membrane [2].

Actinomyces species are normal commensals of the oral cavity, and are also seen in gastrointestinal, respiratory and female genital tracts. The bacteria are opportunistic and cause lesions only when the epithelial defences are weak [2]. Actinomyces act like an intracellular parasites, resist phagocytosis and spread to adjacent tissues without any barriers [3].

Tonsillar actinomycosis is usually a bilateral feature [4]. Although actinomycosis can be detected in the tonsils comparatively frequently, there are only few reports regarding unilateral tonsillar hypertrophy due to actinomycosis. To our knowledge, only six cases of unilateral tonsillar hypertrophy caused by Actinomyces infection have been reported until now. Four of them were masquerading as a tumor, and two as pendulous masses attached to the tonsil [4-9]. We herein report, a case of unilateral Actinomycosis in an 18 year old boy masquerading as a tonsillar neoplasm, as the seventh ever reported case of unilateral tonsillar actinomycosis.

Case Presentation

An 18-year-old boy presented to our outpatient clinic with difficulty in breathing and swallowing along with increased frequency of episodes of sore throat for 3 months. On examination the patient was undernourished. Pharyngeal examination showed ulcero-proliferative growth of the left tonsil measuring 4.5 cm x 3.0 cm. Other tonsil appeared normal. Neck examination revealed a single, smooth, firm, non-tender 2 cm x 2cm palpable cervical lymph node. It was subjected to fine needle aspiration cytology, which showed features of suppurative lymphadenopathy. Ear, laryngeal, nasopharyngeal, per abdomen and cardiovascular examinations were normal. Haematological parameters were within normal limits. Throat culture was negative. Based on clinical findings, a possible diagnosis of tonsillar neoplasm was made.

The patient underwent bilateral tonsillectomy under general anaesthesia. The dissected tonsils were labelled and sent separately for histopathological examination. Hematoxylin-eosin (H&E)-stained tissue sections of the left tonsil demonstrated "sulfur granules" of actinomyces in dilated crypts (Figure 1) and features of reactive lymphoid follicular hyperplasia. On PAS staining, the specimen was found to be positive for typical sulfur granules, and aggregates of filamentous microorganisms were seen (Figure 2). No evidence of malignancy was found. The right tonsil demonstrated follicular hyperplasia without any foci of actinomyces infection. A diagnosis of unilateral actinomycosis was made on histopathology.

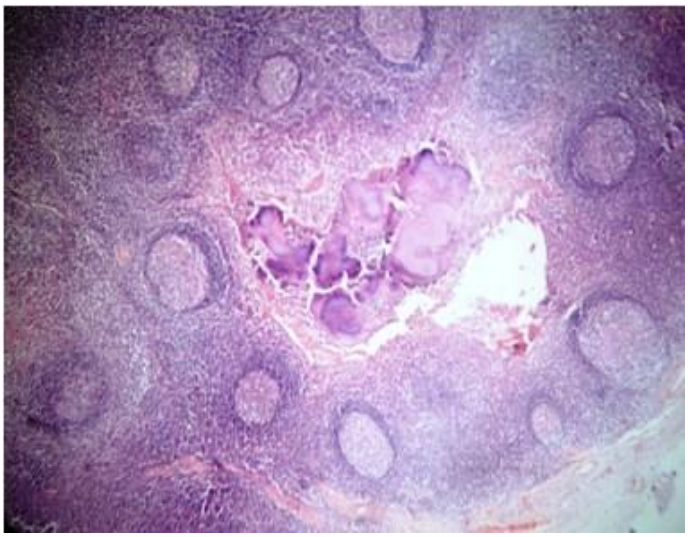

Figure 1 Low power showing "sulfur granules" of Actinomyces inside the tonsillar crypt and hyperplastic lymphoid follicles (H/E, 10X).

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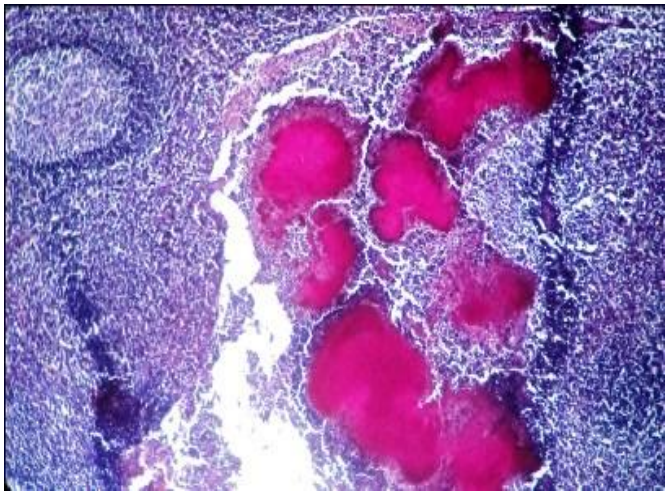

Figure 2 High power view showing PAS positive actinomyces colony (PAS, 40X).

The postoperative period was uneventful. Patient is under follow-up and is keeping well, without any signs of recurrence.

Discussion

The first reported case of human infection with actinomycosis was reported by Von Langenbeck in 1845 and was attributed to a fungus [10]. Bollinger in 1877 first used the term "actinomycosis" with reference to a disease of cattle characterized by a woody swelling of the tongue and diffuse enlargement of the jaw and named it as "lumpy jaw disease" [11]. In the same year later, Harz named the infective organism as 'Actinomyces *bovis*', which meant ray fungus, reflecting that the organism was thought to be a fungus that caused the disease in the cattle [12]. In 1878, James Israel, independently of Bollinger and Harz, demonstrated a similar fungus in a human case[11-13]. The similarity of these findings was pointed out by Ponfick in 1879 and 1882 and he considered the disease to be the same [14]. In 1890, Bostroem gave the first bacteriological description of the organism, and in 1891 Wolff and Israel wrote a description of the pathology based upon two human cases; they isolated an anaerobic form which grew at body temperature, and showed true branching [15,16].

Actinomycosis is characterized by the formation of slightly painful pseudotumours, of slow growth and which may progress into abscesses and form fistulous tracts. In 55% of cases they are located in the cervicofacial region; in 20%, thoracic; and in 15%, abdomino-pelvic. When actinomyces occurs in atypical locations (base of tongue, larynx, central nervous system, bone) they are difficult to diagnose as they tend to simulate malignancies [1]. Frequency of actinomyces in tonsils is 6.7 % to 28.5 % [4].

Tonsillar actinomycosis is usually a bilateral feature. In the differential diagnosis, when one tonsil is much larger than the other, the clinician must suspect unusual infections (botryomycosis, nocardiasis) or neoplasia as the etiology. Mycobacterium tuberculosis, atypical mycobacterium, fungal organism or actinomycosis may all be infections causing unilateral hypertrophy [17]. In our case, ZN stain and PAS stain ruled out mycobacterium tuberculosis and fungal infection respectively. Bhargava *et al* showed a significant association of actinomycosis and tonsillar hypertrophy [18]. According to van Lierop *et al*, there was no correlation found between the presence of tonsillar actinomycosis and recurrent tonsillitis and/or obstructive tonsillar hypertrophy [17],. Aydin *et al* found that the rate of actinomycosis was significantly higher in adults than in children [19]. Since cryptitis (inflammation of tonsillar crypts) is often associated with actinomycosis, it can be considered as a histopathological indicator for tonsillar actinomycosis [19].

For the establishment of diagnosis of actinomycosis, following two conditions are must- positive cultures, sulfur granules or biopsy specimens showing organisms [20]. In our patient, biopsy specimens showed "sulfur granules" of actinomyces in dilated tonsillar crypts which were positive for PAS stain.

The present case is being presented because clinically the patient was undernourished and on pharyngeal examination an ulcero-proliferative growth of the left tonsil was noted which bleeds on touch, favouring diagnosis of tonsillar neoplasm causing a considerable diagnostic dilemma.

Conclusion

The possibility of actinomycosis as a differential diagnosis of a unilateral tonsillar mass should be ruled out before attributing it to some other etiology. Prompt clinical suspicion is needed to diagnose and a long term therapy is needed to successfully treat the infection.

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Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

Authors' Contributions

MKU participated in the clinico-histopathological diagnosis, data acquisition, literature search, drafting, reviewing and editing of the manuscript. PRM, YBR and RR participated in the clinico-histopathological diagnosis, reviewing and editing of the manuscript. All the authors read and approved the final manuscript.

Competing Interests

The authors declare that they have no competing interests.

Funding

Sources of funding- None

Please cite this paper as: Usha MK, Madhavrao PR, Ramling YB, Royal R. Unilateral tonsillar actinomycosis masquerading as a neoplasm. *Int J Stud Res* 2012;2(1):36-8. doi: http://dx.doi.org/10.5549/IJSR.2.1.36-38

Received: 4 Feb 2012, Accepted: 27 Mar 2012