Uncommon Presentation of a Rare Cases- Incidental Finding in a Tonsillectomy Specimen in Mosul City: Histopathological Study

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ABSTRACT:

BACKGROUND:

Histopathological examination in a patient who did tonsillectomy due to various indications reveals some of a rare but important incidental pathological finding as choriostomatic tissue (cartilage and fat) and an unusual bacterial colonization (actinomycosis) in tonsillar specimen.

OBJECTIVE:

The aim of this study is to represent of an unusual finding of choriostomatic tissue in resected tonsils, and to evaluate the incidence of Actinomyces colonization with disclose their relation to tonsillar diseases.

PATIENTS AND METHODS:

A case series study design is adopted. The sample of the present study included 100 patients who have done tonsillectomy are carried out in the ENT Department of Al-jamhori Teaching Hospital in Mosul city during the period from February 2018 to February 2019.

RESULTS:

In the current study, the most frequent indication for tonsillectomy is recurrent tonsilitis. Also shows that there is highly significant relation between incidence of actinomycosis with chronic, recurrent tonsillitis and hypertrophied tonsils (p=0.000, 0.000, 0.003) respectively, there was a statistically insignificant relationship between the presence of actinomycosis and age furthermore there is predominance of actinomycosis in tonsillar obstructive hypertrophy 4 (18.2%), moreover their occurrence in adult patient is more common than in children. The mesenchyme cartilage is seen only in two adult cases (male 2.3%, female, 2.9%) has recurrent and chronic tonsillitis, respectively.

CONCLUSION:

A histopathological analysis of the tonsillar specimen aids in the diagnosis of actinomycosis organisms which are difficult defined in culture. Although choristoma in tonsillectomy specimen is uncommon entity. However, any patient with recurrent or chronic tonsillitis, suspicion for this condition should be considered.

KEYWORDS Tonsillitis, Actinomycosis, Chriostoma

INTRODUCTION:

Palatine tonsil is a part of Waldeyer's lymphoid tissue ring. [1] It has a considerable role in the immune defense mechanism of the body [2]. The most common indications for tonsillectomy are infection and obstruction. Previously, infection was the main indication; while, recently obstruction is regarded to be more frequent indication [3]. Although most frequently operation done in Iraqi children is tonsillectomy, the routine histopathological examination of the tonsillectomy specimen is not recommended except in patients having a high-risk factor for malignancy [4]. Even supposing the tonsillitis is reported as a non-serious disease, but a rare incidental finding of tonsillectomy specimen may alter the methods of management, however, an unusual finding, such as bacterial colony (actinomycosis) or heterotopic tissue (choristoma) may have contribution for a better understanding of the pathogenesis and management of tonsillar disease [5].

Choristoma is a mass of normal tissue that aggregate in an abnormal anatomical site [5]. They can be cartilage, bone, and others [6]. A limited number of tonsillar choristomal tissue has been recorded till now [7], moreover, patients with tonsillar choristomatic tissue, have more possibility to find second cartilaginous tissue in a another places in their body [8], although the surgical excision of choristomatic tissue have a good prognosis and its treatment of choice

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but recurrence have been reported [8] Moreover Actinomycosis is a rare, infectious disease, it has been reported by many authors in a variable percentage in tonsillar specimens [9] in addition it has been reported that actinomycosis has an important role in tonsillar diseases [10]. Despite of the exact mechanism of this pathogen to cause disease is not clearly understood [11], actinomycetes release proteolytic enzymes which reduces oxidative reduction potential and leads to proliferation of the organisms that invades the surrounding tissues [12, 13]. Actinomyces are anaerobic, gram positive, bacteria, found in the oral cavity [14].

The aim of present study is to assess the actinomyces colonization in patients undergoing tonsillectomy in and to disclose any possible role of Actinomyces in a tonsillar disease in Mosul city. In addition to report the incidence of unusual choristomatic tissue (cartilage, fat) in tonsillectomy specimen.

PATIENT AND METHODS:

A total of 100 patients who have done tonsillectomy due to obstructive hypertrophied tonsils and/or infectious diseases in form of series study design. This study was carried out in the ENT department of Al-Jamhori Teaching Hospital in Mosul city during the period from February 2018 to February 2019. Informed consent was taken from all patients. The diagnosis of tonsillar diseases was done by ENT specialist. Tonsillar specimens are processed to get paraffin blocks, next the section is stained with hematoxylin and eosin and examined under light microscopy with resolution power 10x and 40× (Olympus BX 51; Tokyo, Japan). Reports are noted and the data were analyzed by using SPSS version [12] .

RESULT:

This study recorded that the *most frequent indication* for tonsillectomy is *recurrent tonsilitis* in 44% of patient, while 34% where due to chronic tonsillitis and 22% of the *patients* have obstructive hypertrophied tonsils as in **figure 1**.

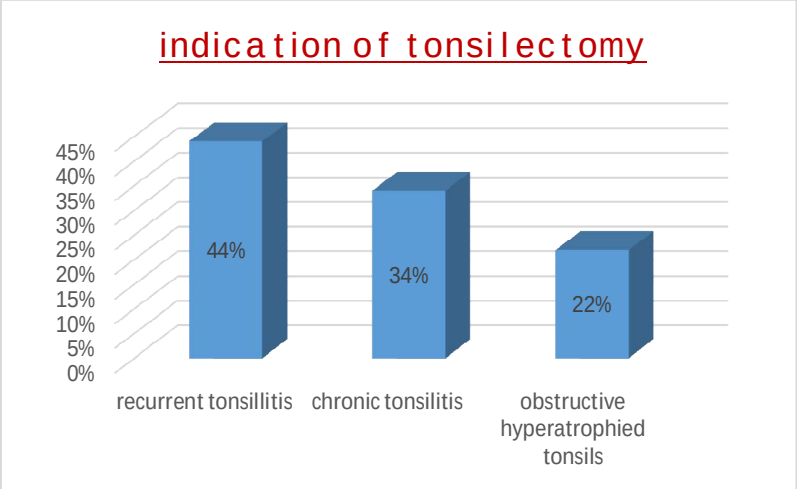

Figure (1): Indication of tonsillectomy

Table (1) shows that there is highly significant relation between incidence of actinomycosis with chronic, recurrent tonsillitis and hypertrophied tonsils (p=0.000, 0.000, 0.003) respectively, there was a statistically insignificant relationship between the presence of actinomycosis and age (p=0.125) moreover the actinomycosis being more common in older patients. There is predominance of this microorganism in 4 (18.2) of hypertrophied tonsills. While in recurrent tonsillitis, 4/34 patients (11.7%) have actinomyces colonies. In addition, actinomycosis is detected in 6 cases 13.6% who did tonsillectomy due to chronic tonsillitis (Figure 2, 3)

Table 1: Incidence of actinomycosis according to indication of surgery and age grouping

Group	Total (n=100)	No. of patients with actinomycosis	P*- value
Chronic	44	6 (13.6)	0.000
Recurrent	34	4 (11.7)	0.000
Hypertrophic	22	4 (18.2)	0.003
P**- value	0.792		
Age			
≤ 20 year	85	10 (11.8)	***0.125
> 20 year	15	4 (26.6)	

*Z test of one proportion
** One-way ANOVA test & post hoc (Duncan) Test was used.
***Chi-square test was used

The presence of Mesenchymal choristomatic tissue

All sections were examined histopathologically to see the presence of rare mesenchymal tissue. The results show that mesenchymal choristomatic tissue (cartilage, fat, bone) is very limited and rare in tonsillectomy specimens. There is no significant correlation between the presence of choristomatic tissue and tonsillar diseases (p=0.627).

The mesenchymal cartilage is seen only in two adult cases (male 2.3%, female, 2.9%) who had recurrent and chronic tonsillitis, respectively. On the other hand, one case (2.9%) is seen with fat mesenchyme in the chronic tonsillitis group (Figures 4, 5, 6).

Table 2: Mesenchymal choristomatic tissue in the palatine tonsillectomy specimen

Out of 100 patients		Recurrent tonsillitis (n=44)	Chronic tonsillitis (n=34)	Hypertrophic tonsils (n=22)	P* - value
The presence of Mesenchymal choristomatic tissue	Absent	43(97.7%)	32(94.2%)	22(100%)	0.627
	Fat	0	1(2.9%)	0	
	Bone	0	0	0	
	Cartilage	2 (2,3%)	1(2.9%)	0	

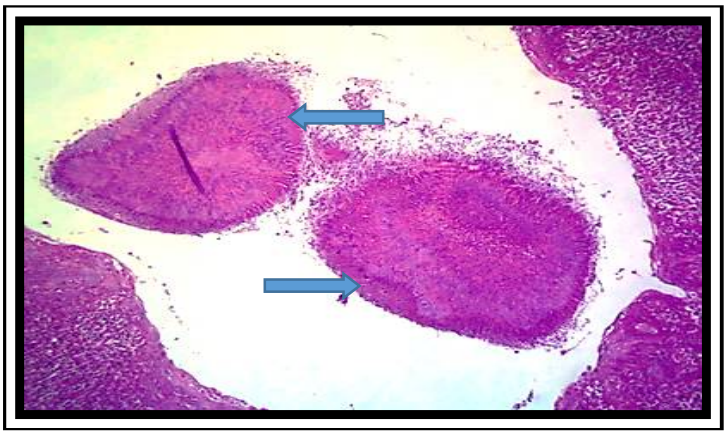

Figure 2: Histopathologic section of tonsil demonstrating a tonsillar actinomycosis (arrow). H&E, X100

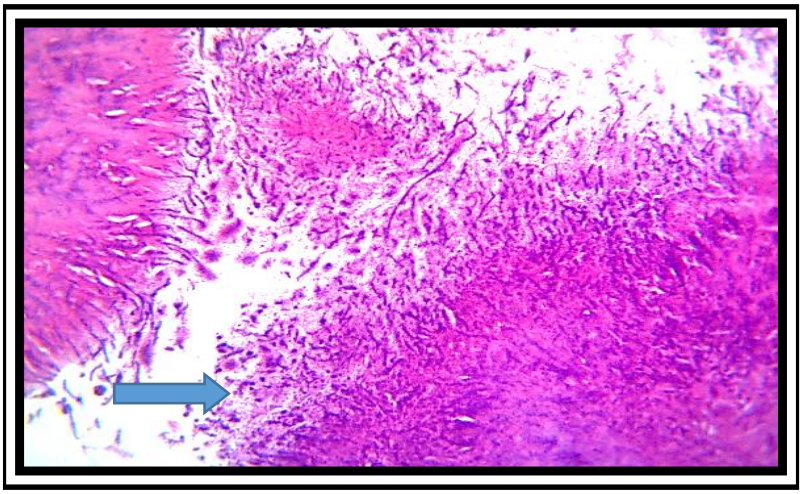

Figure 3: Tonsil showing actinomycotic bacterial colony within the tonsillar crypt (arrow). H&E, X400

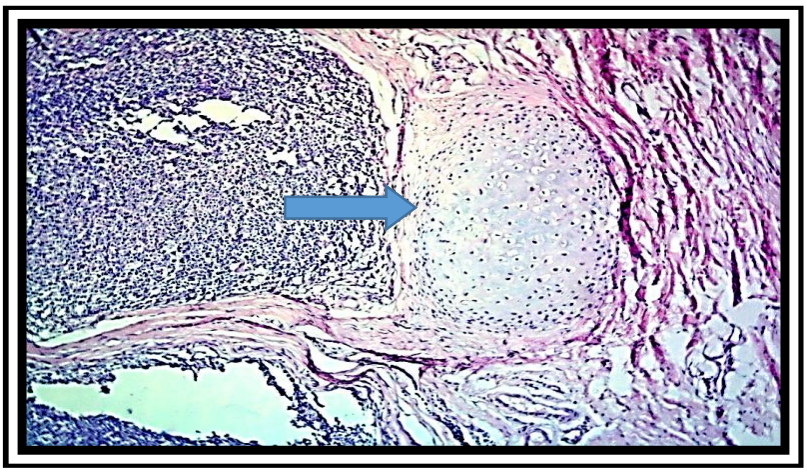

Figure 4: Histopathology section in tonsil of male patient 21years old showing islands of cartilage adjacent to follicular hyperplasia in tonsil (arrow). H&E, X100.

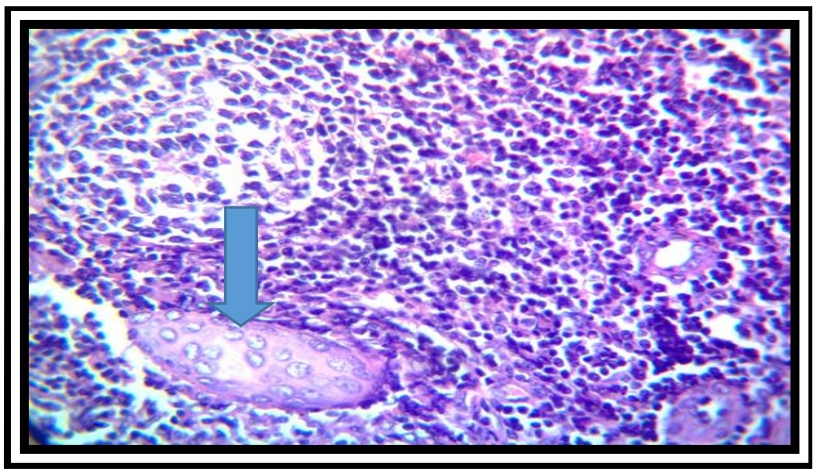

Figure 5: Histopathology section in tonsil of female patient 15 years old showing islands of hyaline cartilage adjacent to follicular hyperplasia in tonsil (arrow). H&E, X100.

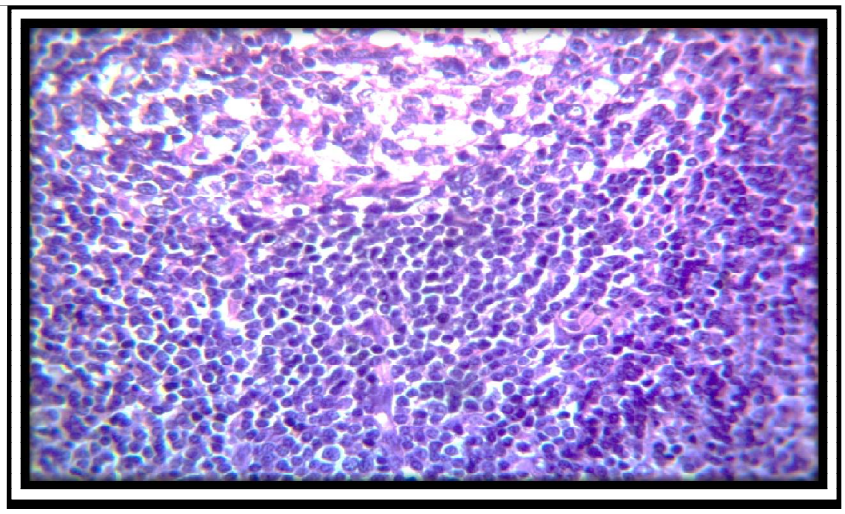

Figure 6: Microscopic section in male tonsil: showed fat choristoma seen within lymphoid (arrow). H&E, X100

DISCUSSION:

Although actinomycotic is unusual infections of the head and neck region but it represents an important entity because of its varied presentation, difficult diagnosis, and long course of treatment. In addition, it can mimic oropharyngeal malignancies [15]. From one hundred patient which are contributed in this study, 14% were positive for bacterial colonies actinomyces with insignificant p value =0.792, in spite of predominance of this species in hypertrophic tonsils compared to the other tonsillar diseases.

Although the relation ship of actinomycosis and tonsillar diseases has been reported in many previous studies [16,17] but this has been a point of contention between various authors, Ashraf study (2011) considered the prevalence rate of tonsillar actinomycetes colonization is lower in patients who have obstructive tonsillar hypertrophy 26.3% than in patients who have recurrent tonsillitis as indication for tonsillectomy 43.9%, while others considered bacterial colonies higher in patients who did tonsillectomy for sleep-disordered breathing [18,19]. Kansu *et al.*, (2017) and Bhargava *et al.*, (2001) foun d similar a positive correlation between actinomycosis and tonsillar hypertrophy; where actinomycosis is present in 56.8% of patients with tonsillar hypertrophy compared to 10.3% in patients with recurrent ton sillitis [20, 21].

However, the current results are inconsistent with previous studies which reported that there is no statistical significance correlation between the presence of actinomycosis and the tonsillar disease [22, 23, and 24].

There is controversial regarding the incidence of actinomycotic clumps in tonsillectomy specimens in the available review of literature, Furthermore the incidence of occurrence of this microorganism are seen between 2-30% [18, 19, and 23]. Aydın *et al.* (2005), how examine 1820 tonsillectomy specimens in retrospective study and recorded the incidence of Actinomyces to be 6.7% [22].On the other hand a new study by Maharjan *et al.*, 2017 recorded an incidence of actinomycosis to be as high as 56% of the total cases, the result above agree with current study in that there is a significantly higher incidence of actinomyces in adult tonsillar tissue compared with pediatric population [18, 20, 16].

This difference may be explained by attributed to the method used in the study, environmental, regional differences, cultural factors [16] Some authors recommend that persistent stimulation by actinomycosis may explained the development of hypertrophy [26, 14]. For this reason, researchers recommended to try oral penicillin in patient with obstructive sleep apnea to eradicate the organism and diminish tonsillar size [16].

Histopathology is the most accurate diagnostic procedure to find tonsillar actinomycosis in stead of culturing it [16]. One of the most interesting studies about the possible cause of the increase's incident of Actinomyces in hypertrophied tonsil might be the presence of subclinical colonization of bacterial colonies which activates and stimulates lymphoid hyperplasia, leading to tonsillar hyperplasia [27]. Ozgursoy *et al*. (2008) reported that actinomycosis infection is able to trigger the mechanism for tonsillar hypertrophied lymphoid tissue.

The presence of Mesenchymal choristomatic tissue:

Although choristomas are rare entity of academic interest, and only few systematic studies reported the presence of choristomatic tissue are noted till now [28]. However, the present study is based on the presence of cartilage, fat and bone mesenchymal tissue in tonsillectomy specimen, the results show that there is a limitation in the existence of choristomatic tissue in tonsillectomy specimen; this agree with many studies [29,30]. Moreover, in Turkey, Abdulla *et al.*, (2002) estimated that choristomatic tissue was (3%,1%, 1%) as cartilage, fat, and bone respectively [29]. While a study achieved by Bharti et al 2013 in India declared that mesenchymal tissue (cartilage, fat, and bone) was (9.4, 3.5 and 1.17 percentage) correspondingly in tonsillectomy specimen [30]. A chondroid choristoma is first described by Berry in 1890, as one of the causes of recurrent tonsillitis, and pathologists must be aware and not to be confused with the neoplasms [30].

Some authors proposed mechanism for the development of cartilage proliferation and bone formation in chronic inflammation that choristoma of tonsils occurs as a developmental disorder leads to the development of aberrant mesenchymal tissue [29] Other mechanism is the ability of mesenchymal progenitor cells to differentiate into various mesenchymal lineages or production of osteogenic substances [5].

The cartilaginous metaplasia is a differential diagnosis of cartilaginous choristomas, is often associated with scattered cartilaginous cells and ill-fitting dentures arranged in various stages of maturation (Sulhyan *et al*., 2016; Jyotsna *et al*., 2018) although tonsillar cartilaginous choristoma is a rare entity and includes a very small percentage of all masses in the nasopharynx, there is a possibility to find another cartilage in a different place in the body [5, 31].

CONCLUSION:

A histopathological analysis of the tonsillar specimen aids in the exact diagnosis of actinomycosis organisms which are difficult to culture. Highly significant correlation was found between the presence of tonsillar actinomycosis and recurrent tonsillitis, chronic and obstructive tonsillar hypertrophy Histopathologic findings showed presence of Actinomyces as colonization reaction in the tonsils. There was significant correlation between actinomycosis colonization and age that actinomycosis being more common in older patients. Choristoma in tonsillectomy specimen is uncommon entity. However, any patient with recurrent or chronic tonsillitis, suspicion for this condition should be considered.

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