



# Occult malignancy in pediatric tonsil and adenoid surgeries – A national survey<sup>☆</sup>

Guy Lifshitz<sup>a</sup>, Maayan Gruber<sup>a,b</sup>, Ohad Ronen<sup>a,b,\*</sup>

<sup>a</sup> Azrieli Faculty of Medicine, Bar Ilan University, Safed, Israel

<sup>b</sup> Department of Otolaryngology – Head and Neck Surgery, Galilee Medical Center, Nahariya, Israel



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## ABSTRACT

**Objective:** To evaluate the incidence of tonsil and adenoid malignancy in pediatric patients in Israel compared to the known literature and to revisit the common practice of routine histopathologic examination of tonsils and adenoids.

**Methods:** Analysis of the Israel National Cancer Registry data on pediatric tonsil and adenoid malignancies between the years 2005 and 2015, and a systematic literature review of all relevant articles that reported on malignancies amongst pediatric patients who underwent tonsillectomy with or without adenoidectomy.

**Results:** Only seven cases of tonsillar malignancies were documented out of 152,352 (0.0052%) surgeries in the pediatric population. All malignancies were lymphoproliferative and mainly Burkitt's lymphoma. In our medical center no malignancy was found in 2165 patients in the same age groups and time period. We found the incidence of tonsillar malignancy in Israel to be significantly lower ( $p = 0.013$ ) compared to the previously described literature.

**Conclusion:** The incidence of malignancy in routine histopathological examination of tonsils and adenoids in Israel is very low when compared to the known literature. According to these results, we suggest that routine histopathological examination of all such samples is not clinically justified.

## 1. Introduction

Tonsillectomy and adenoidectomy is one of the most common surgical procedures performed in children worldwide [1]. In Israel alone, 10,000–17,000 such surgeries are performed every year, and more than 500,000 each year in USA [2]. The common indications for tonsillectomy with or without adenoidectomy can be divided into two groups [3]: recurrent or chronic infections or sleep disordered breathing. Absolute indication for removal of tonsils/adenoids is to rule out malignancies in suspicious cases. Lymphomas are the most common neoplasms of the head and neck in children although other pathologies include soft tissue sarcomas, neural tumors and thyroid malignancies [4,5]. Until recently, tonsils and adenoids were sent to routine histopathologic examination as a common practice. In recent years, several studies have questioned whether this routine examination is clinically justified in the pediatric population. In a wide scale literature review of pediatric tonsillectomies, Randell et al. [2] found only 6 tonsillar malignancies out of 13,547 specimens (0.044%). Out of these 6 malignancies, 2 patients presented with no signs or symptoms for neoplasm

disease. In another review of children under 16 years old, Garvallo et al. [6] reported only 2 cases of tonsillar malignancy out of 1123 cases. Verma et al. [3] found no malignancy in 1017 children who had undergone tonsillectomy in their hospital. Dohar et al. [7] reported 5 years' experience at Children's Hospital in Pittsburgh in which there was only one case of suspected tonsillar lymphoma out of 2012 children. Considering the above literature, the aim of this study was to calculate the incidence of tonsillar and adenoid malignancy in Israel based on the Israel National Cancer Registry database, and to evaluate whether routine histopathological examination is clinically justified.

## 2. Materials and methods

This retrospective data review examined records from the Department of Otolaryngology – Head and Neck Surgery and the Department of Pathology at Galilee Medical Center, the Israel Ministry of Health, and the Israel National Cancer Registry.

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\* Corresponding author. Galilee Medical Center, POB 21, Nahariya, 2210001, Israel.

E-mail address: [ohadr@gmc.gov.il](mailto:ohadr@gmc.gov.il) (O. Ronen).

## 2.1. Population

We included all patients under 18 years old who underwent tonsillectomy with or without adenoidectomy for any indication in Israel between 2005 and 2015 by correlated ICD-9 procedure codes. A PubMed literature review of all the relevant studies that examined the incidence of tonsil and adenoid malignancy in a pediatric population was done. Exclusion criteria included: patients older than 18 years, case reports, and non-English-language studies. Children who presented with suspected malignancy, tonsillar asymmetry or cervical lymphadenopathy were not excluded from the cohort.

## 2.2. Statistical analysis

For statistical analysis we used Fisher's exact test and Pearson  $\chi^2$  [2] test to compare between the national and international results. A  $p$  value less than 0.05 was considered statistically significant.

## 3. Results

**National data:** We found 152,352 procedures of tonsillectomy and adenoidectomy performed between 2005 and 2015 in Israel. There was a steady increase in the number of surgeries in recent years (Fig. 1). Since 2005, significant elevation is seen with a peak incidence in 2013 and since then a slight decrease in 2014–2015. The different procedures breakdown according to the different ICD-9 codes is presented in Fig. 2. The majority of surgeries (58%) are adenoidectomy alone, followed by tonsillectomy & adenoidectomy (24%), and tonsillectomy alone (18%) of all surgeries. Overall, between 2005 and 2015, eight cases of tonsillar malignancies were found throughout Israel in children under 18 years old. No patients with malignancy were found in the database of the Galilee Medical Center. All neoplasms were lymphomas, seven of them classified as Burkitt's lymphoma and one as diffuse large B cell lymphoma. The distribution of tonsils neoplasm according to the age groups: one case in ages 0–4, five in ages 5–9, none in ages 10–14, and one in ages 15–18.

**World data:** The systematic literature review conducted in PubMed found 194 related articles using the key words: 'Tonsils'; 'Adenoid'; 'Children' or 'Pediatric'; 'Malignancy' or 'Lymphoma'. After implementing the inclusion criteria of age 0–18, tonsillectomy and/or adenoidectomy, prospective, cohort, or retrospective studies, and a diagnosis of malignancy, only 15 relevant papers were found (see Table 1). After excluding duplicate and unnecessary data we summarized 30,724 patients under 18 years old who underwent tonsillectomy with or without adenoidectomy and whose specimens were examined. Out of these studies only 10 cases (0.032%) of tonsillar neoplasm, including one synchronous tonsillar adenoid malignancy, were diagnosed by histologic exam, as can be seen in Table 1. Overall, four cases (40%) were diagnosed with Burkitt's lymphoma in patients aged 2, 5, 6, and 8 years old. Another three (30%) patients were tagged as unspecified lymphoma due to lack of information, and there was one case each

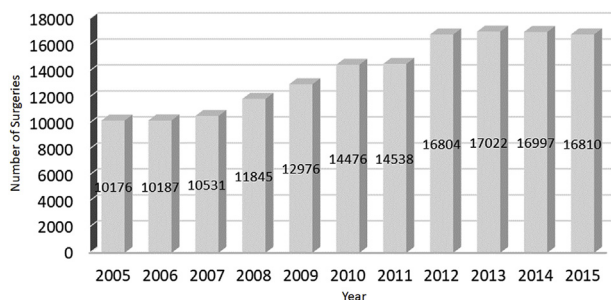


Fig. 1. Graphic distribution of tonsillectomy & adenoidectomy of the pediatric population in Israel per year.

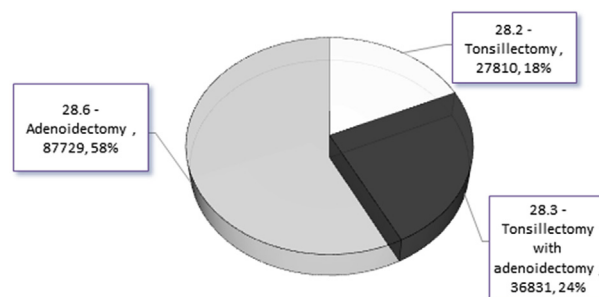


Fig. 2. Breakdown of each surgery by ICD-9 code between the mentioned years. In rectangles – ICD-9 code, name of procedure, total number of surgeries performed, and its percentage out of the total surgeries performed.

(10%) of T-cell lymphoblastic lymphoma, DLBCL, and follicular lymphoma.

**Comparison between national and international data:** The incidence of tonsillar neoplasm in pediatric patients in Israel (0.0052%) was found to be significantly lower ( $p < 0.0001$ ) than the findings in our literature review (0.032%). The incidence of tonsillar malignancy per 10,000 children was 0.52 in Israel compared to 3.2 in studies from other countries. The peak incidence of tonsillar neoplasm was found in the 5 to 9-year-old age group both in Israel and the world (Fig. 3). Unfortunately, the age grouping division system of the Israel National Cancer Registry prevents us from comparing the neoplasm incidence more exactly within that age group.

## 4. Discussion

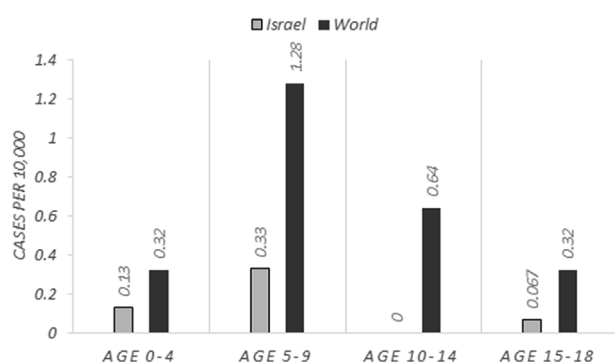
This study presents the largest published cohort of pediatric adeno-tonsillar surgeries correlated with histopathological reports. Combining the numbers of all the published series in the English literature so far yields 30,724 compared to 152,352 cases in this report. A total of 10 malignancies were reported throughout the combined previous studies compared to eight cases in this presented cohort.

The overall characteristics and features of tonsillar neoplasm such as peak age and type of lymphoma in our national database were found to be generally similar to those in the professional literature. Our aim was to evaluate the incidence of occult malignancies out of all tonsillar neoplasms in order to stratify the optional risk of not performing routine histological examination on the specimens. We were able to locate the clinical records of six of the eight children who were diagnosed with malignancies in Israel. All six cases of tonsillar neoplasm presented with pre-operative high index of suspicion and were considered “absolute” indication for surgery. Our national survey includes high-volume data from national records, which enabled us to compare it to previous international studies that described one center's experience or retrospective review. On the other hand, disadvantages were lack of information about patients' clinical presentation and manifestation and the inability to assess the number of specimens not sent to microscopic evaluation out of the 152,352 surgeries. We found several centers that did not send the pathology specimens of tonsils and adenoids for pathology evaluation. Moreover, in recent years some novel approaches to adenoidectomy and partial tonsillectomy have become more acceptable. Methods such as ‘coblation tonsillotomy’ do not produce any pathological specimen, potentially reducing the number of relevant pathology results.

Two studies, one by Yalon et al. [8] and a more recent study by Pham et al. [9], found that the vast majority of pediatric patients with tonsil asymmetry were not diagnosed with lymphoma in final histological analysis. These conclusions are in line with the fact that six out of eight Israeli patients who were diagnosed with lymphoma in our cohort presented with high malignancy suspicion prior to surgery. Therefore, we can carefully assume that the remaining two patients (25%) may be

**Table 1**  
PubMed reviews of tonsils and adenoid malignancies.

Author	No. Patients	No. Malignancies	Type of Malignancy	Age	Site	Clinical Signs
Ridgway, 1987 [10]	1100	0				
Dohar, 1996 [7]	2012	1	Lymphoma Unspecified	Unknown	Tonsil	Asymmetric tonsil
Strong, 2001 [11]	1539	0				
Younis, 2001 [12]	2099	0				
Williams, 2003 [13]	4070	2	1 Burkitt's Lymphoma 1 Lymphoma Unspecified	2 Y/O 11 Y/O	Tonsil Tonsil	Necrotic tonsil Cervical adenopathy
Garavello, 2003 [6]	1123	2	2 Burkitt's Lymphoma	6 Y/O 8 Y/O	Tonsil Tonsil	None None
Erdag, 2005 [1]	2743	0				
Dost, 2006 [14]	400	0				
Yasan, 2006 [15]	1216	0				
Dewil, 2006 [16]	2058	0				
Verma, 2009 [3]	2062	0				
Papouliakos, 2009 [17]	753	1	1 Unspecified Non-Hodgkin Lymphoma	6 Y/O	Tonsil	Asymmetric tonsil
Van Lierop 2009 [18]	172	1	T Lymphoblastic Lymphoma	Unknown	Tonsil & Adenoid	B symptoms & Cervical adenopathy
Booth, 2013 [19]	570	3	1 Burkitt's Lymphoma 1 Follicular Lymphoma 1 DLCL	5 Y/O 14 Y/O 15 Y/O		
Bizzell, 2017 [20]	8807	0				
<b>Total</b>	<b>30,724</b>	<b>10</b>	<b>4 Burkitt's Lymphoma</b> <b>3 Lymphoma Unspecified</b> <b>1 T Lymphoblastic Lymphoma</b> <b>1 DLCL</b> <b>1 Follicular Lymphoma</b>		<b>10 Tonsils</b> <b>1 Adenoid</b>	<b>6/10 (60%)</b>



**Fig. 3.** Tonsil & adenoid malignancies per 10,000 specimens in Israel compared to the collected literature data. Grey column depicts current study findings in Israel, black columns shows the averages found in world literature.

classified as having an occult malignancy.

There is no explanation in the published literature for the occurrence of occult disease in pediatric tonsils and adenoid [1,3,6,7,10–20]. A simple explanation might be that the surgery is one of the most common procedures done in children for other reasons, such as obstructive sleep apnea and recurrent tonsillitis and that in many centers the tonsils and adenoids are routinely sent for pathology. It is yet to be investigated whether the early incidental diagnosis of a lymphoma in tonsils and adenoids in the pediatric population is related to better prognosis.

When considering the low incidence of tonsillar malignancy amongst children and the additional monetary expense, estimated to be around \$700,000 each year in Israel, we believe that routine examinations of tonsil and adenoid specimens in the pediatric population might not be justified.

## 5. Conclusions

In this study we report the largest published cohort of pediatric adeno-tonsillar surgeries correlated with histopathological reports. We found that the incidence of tonsil neoplasm in pediatric patients in Israel is significantly lower compared to the previously published data. In most adeno-tonsillar lymphoma cases, alarming signs and symptoms

were presented before surgery, and a pathology evaluation in these cases was obviously warranted. According to the above data, we suggest that routine histopathological examinations are not clinically justified in pediatric adeno-tonsillar surgeries.

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## Conflicts of interest

The authors have no conflicts of interest to disclose.

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