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2022

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How to cite

TROTTET, Lucille et al. A Retrospective Clinico-Pathological Analysis with Review of Literature of Oral and Cervical Lympho-Epithelial Cysts from a Pathological Perspective. In: Applied sciences, 2022, vol. 12, n° 5, p. 2525. doi: 10.3390/app12052525

This publication URL: <https://archive-ouverte.unige.ch/unige:162713>

Publication DOI: [10.3390/app12052525](https://doi.org/10.3390/app12052525)

Article

A Retrospective Clinico-Pathological Analysis with Review of Literature of Oral and Cervical Lympho-Epithelial Cysts from a Pathological Perspective

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Featured Application: Overview of lympho-epithelial cysts.

Abstract: Oral and cervical lympho-epithelial cysts (LECs) are uncommon lesions with histopathological similarities. The aim of the study is to present clinico-pathological characteristics of oral and cervical LECs with a review of literature in order to create awareness on this uncommon entity. Eighteen new cases of oral and cervical LECs obtained from the archives of the University of Peradeniya and University of Geneva were used for the clinico-pathological analysis. The average age at diagnosis of 7 oral and 11 cervical LECs were 40 and 36 years, respectively. Both showed a female predilection (male:female ratio at 3:4 and 4:7, respectively). The only difference was found in the size of the lesions with oral LECs being significantly smaller than cervical LECs (0.9 cm vs. 4.6 cm). LECs may clinically resemble neoplasms (4/18), including malignancies (1/11 in the present series). None of the 18 LECs recurred following surgical removal. The literature-review-based analysis of 514 oral LECs confirms that the lesions are observed predominantly in adults in 4th–5th decades of life and are relatively small lesions of less than 2 cm in diameter. Oral LECs were found to occur predominantly in the tongue and floor of the mouth, similar to 88% of lesions in literature. In conclusion, oral and cervical LECs are two histologically similar cysts that occur in two distinct sites. The literature review supports the information observed in our study with respect to age of occurrence, site predilections, and size. Cervical LECs, particularly the ones that occur in the parotid region, may require further investigations to exclude BLEL of parotid gland, which occur in HIV infected patients.

Keywords: lympho-epithelial cyst; branchial cyst; demographics; germinal centers



Citation: Trottet, L.; Jayasooriya, P.R.; Abeyasinghe, U.W.A.M.L.; Mendis, R.B.R.N.; Lombardi, T. A Retrospective Clinico-Pathological Analysis with Review of Literature of Oral and Cervical Lympho-Epithelial Cysts from a Pathological Perspective. *Appl. Sci.* **2022**, *12*, 2525. <https://doi.org/10.3390/app12052525>

Academic Editors: Dorina Lauritano and Andrea Scribante

Received: 31 December 2021

Accepted: 25 February 2022

Published: 28 February 2022

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1. Introduction

The lympho-epithelial cysts (LECs) may occur within the oral cavity or in the cervical region. Irrespective of the site of occurrence, both show similar histopathology [1–4]. The cervical LECs were initially referred to as Branchial cysts or Branchial cleft cysts [5–7]. However, as the origin of these cysts remain unclear, current terms based on referring to site of occurrence and histopathological presentation seem more appropriate.

The oral LEC is a rare asymptomatic lesion that appears as a whitish or yellowish sub-mucosal nodule that is generally smaller than 1 centimeter and mobile on palpation. It is found mainly on the floor of the mouth and the lateral border of the tongue. In addition, it can also develop in various intraoral locations such as the buccal vestibule and also the soft palate [4]. Two main theories have been proposed concerning the pathogenesis of the LEC. Knapp's theory [1] suggests that the obstruction of lymphoid crypts induces dilatation and

cystic proliferation of the epithelium. Buchner and Hansen's [2] and Bhaskar's theories [3] assume that cystic proliferation occurs from epithelial cells that persist in lymphoid tissue during embryogenesis. However, both theories fail to explain how oral LECs occur in sites without lymphoid tissue.

The prevalence of oral LECs is difficult to assess because a large number of cases are probably not histopathologically diagnosed, as they are asymptomatic. According to Yang [4], the age of onset is about 45 years with a female predilection (2:1). The clinical diagnosis of the oral LEC is not obvious. It may be confused with an oral mucocele, a lipoma, and possibly a tumor of the minor salivary glands, but never as a malignancy.

The literature indicates that cervical LECs, on the other hand, appear late in childhood or in young adults, generally as a unilateral asymptomatic mass of slow evolution, localized in the anterior triangle of the neck in the superior third of sternocleidomastoid muscle [5–7]. The hypotheses of etiopathogenesis of the cervical LECs indicate that they may arise as a result of an anomaly of fusion of the branchial arches [5], a cystic change of thymic origin [6], or a cystic change in cervical lymph nodes [7], of which the first origin is more widely accepted [5]. In support of the above conclusion, it can be stated that branchial anomalies are frequent and represent about 17% of the cervical masses in pediatric consultations [8]. These abnormalities include cervical lympho-epithelial cysts, but also sinuses and fistulae.

Regarding gender distribution, Agaton-Bonilla et al. [9] and Ford et al. [10] have observed more cases in women than in men, while Gaszynska et al. [11] observed the opposite. McNealy et al. have observed dominance on the right side [6] and Papadogeorgakis et al. on the left side [12]. Literature reveals a wide range of clinical differential diagnoses for cervical LECs, including infections such as tuberculous lymphadenitis, and benign and malignant neoplasms including lipoma, neurofibroma, haemangioma, lymphangioma, carotid body tumours, metastatic neoplasms, lymphomas, teratoma, and parotid neoplasms [13], which may require different management strategies. The cervical LEC is generally asymptomatic. Nevertheless, during super infection, the patient may complain of dysphagia, dyspnea, cough, and dysphonia, depending on the size and location of the cyst.

Accurate histopathological diagnosis is necessary to ensure proper treatment. The treatment of both oral and cervical LEC is surgical excision [4,9–15]. The aim of the present study is to give a clinico-pathological analysis of 18 new cases of LECs. Our data were compared with those of the literature in order to get a better understanding of the oral and cervical LECs so as to improve the clinical and histopathological diagnoses. The rationale for conducting the study was to highlight the comparison between oral and cervical LEC. None of the clinicopathological studies reviewed contained a comparison between oral and cervical LEC. Authors found it relevant to compare the clinicopathological features of the two entities as these are two lesions that share similar histopathological features but occur at different locations. Comparison was also undertaken to evaluate if there are any striking clinico-pathological differences between the entities other than the location.

2. Materials and Methods

Seven cases of oral LECs were recorded, two at the Unit of Medicine and Oral Pathology at the University Hospitals of Geneva and five at the University of Peradeniya, Sri-Lanka. The 11 cases of cervical LECs come from the University of Peradeniya. Cases were selected on the basis of histological diagnosis. Inclusion criteria were the definitive histological diagnosis of oral or cervical LEC and the adequate clinical information available in respective data bases. The cases were excluded in the event that there was inadequate tissue for new H&E sections.

The preparation of the histological sections was performed with the standard haematoxylin-eosin staining to confirm the diagnoses.

The Ethical Committee of the University Hospitals of Geneva, Geneva, Switzerland, approved the protocol (ID 2017-00015), as well as the ERC of the Faculty of Dental Sciences, Sri-Lanka (ERC/FDC/UOP/1/2016/40).

Clinical and medical history data such as age, sex, location, time since the onset of lesion, size, consistency, and presumptive diagnosis were noted.

The literature review was conducted as follows: an initial search string was carried out to identify articles dealing with LEC with the following keywords: [oral AND lymphoepithelial AND cyst], [intraoral AND lymphoepithelial AND cyst], using several search engines including Google Scholar, Medscape, and Pubmed. Articles published from 1966 to 2021 were included. A total of 154 publications were presented directly with the keywords. Abstracts that were present in English were reviewed. Forty-eight non-relevant articles that dealt with lymphoepithelial carcinoma and lymphoepithelial sialadenitis were excluded initially. Thereafter, lymphoepithelial cysts that were associated with parotid glands and studies that dealt with clinico-pathological presentations of cysts other than LEC were excluded. Finally, a total of 10 articles that analyzed clinico-pathological presentations of at least 05 LEC were identified to present the literature review based presentations of LEC [2–4,16–22]. Four hundred and seventy-nine oral LEC were assessed in these studies. In addition, a further 29 articles describing the clinico-pathological presentations of 35 oral LEC are presented in the Supplementary Materials Table S1 [23–52].

3. Results

Tables 1 and 2 show the clinical data of oral and cervical LEC. Table 3 shows a summary of clinico-pathological information of both LECs. All oral LECs were asymptomatic lesions, while a few cervical LECs (2/11) presented with features of inflammation such as pain. Of them, 43% and 45% of oral and cervical LECs had occurred by the 3rd decade of life, although the average ages of occurrence for the two lesions were 40 and 36 years, respectively. Both lesions showed a female predilection. Oral LECs were present from 1 to 36 months at the time of consultation, while patients with cervical LECs waited between 8 days and 3.5 years to consult a clinician with lesions of 2 to 8 centimeters in size. The oral LECs were nodular, soft, or firm on palpation and varied in size from 0.2 to 1.5 centimeters with an average of 0.9 centimeters and were mostly on the floor of the mouth 42% (3), but also on other sites: ventral surface of the tongue (1), buccal mucosa (1), tonsillar fossa (1), and soft palate (1) (Figure 1).

A statistical analysis with Fisher's exact test considering $p < 0.05$ as significant revealed that there were no significant differences with respect to age, gender, and the time duration taken to seek a consultation when oral and cervical LEC were compared. However, there were significant differences with respect to the size of the lesion with oral LEC always presenting significantly smaller than cervical LEC ($p = 0.000$). Invariably, there were statistically significant differences with respect to the site of occurrence with oral LEC showing predilection to the tongue/floor of the mouth and cervical LEC occurring in the region of the parotid or upper part of the neck ($p = 0.000$).

Table 1. Clinical data of the seven oral LEC.

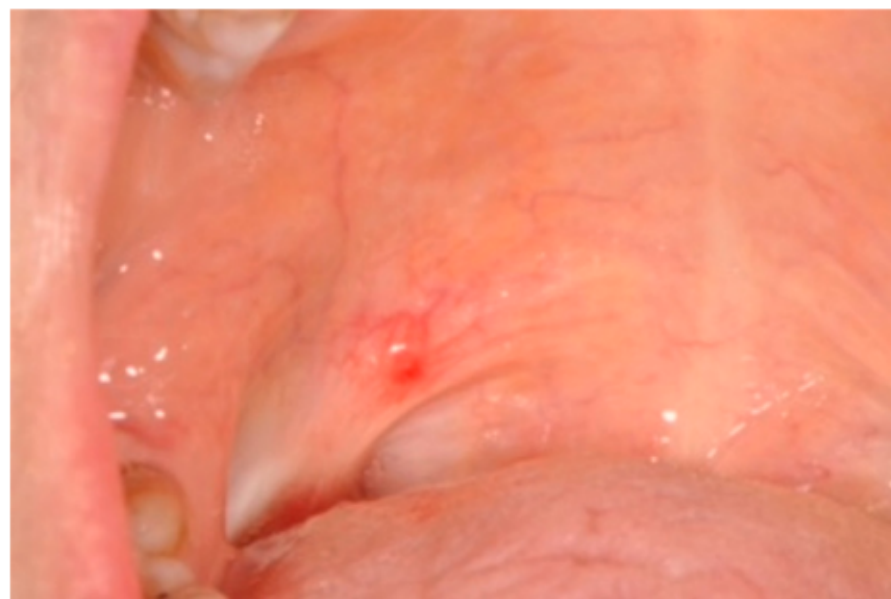
Sex	Age Years	Location	Duration Months	Size cm	Presumptive Diagnosis
M	27	Mouth floor	36	1 × 0.5	Lipoma
M	50	Buccal mucosa	4	1.5 × 1.5	Mucocele
F	68	Tonsillar fossa	8	1.5 × 1.5	Tumour of the accessory salivary glands
F	20	Ventral tongue	2	1 × 1	Fibroma
F	49	Mouth floor	1	0.2 × 0.2	Fibro-epithelial lesion
M	42	Mouth floor	-	0.5	Mucocele
F	26	Soft palate	-	0.6	Nodule

Table 2. Clinical data of the 11 cervical LEC.

Sex	Age Years	Location	Side	Duration Months	Size cm	Presumptive Diagnosis
F	55	Neck close to submandibular region	L	6	-	Infected branchial cyst
F	22	Anterior neck triangle	R	2	4 × 3	Branchial cyst
F	21	Upper part of the neck	L	42	8 × 5	Branchial cyst
F	30	Neck	L	18	6 × 7	Branchial cyst
M	21	Neck	L	1.5	4 × 3	Branchial cyst
F	15	Neck	L	10	3 × 3	-
M	27	Neck	L	6	6 × 5	Branchial cyst/cystic hygroma
F	56	Parotid	R	-	-	Cystic lesion
M	39	Upper part of the neck	L	-	-	Lymphoma, parapharyngeal tumour
M	61	Parotid	R	4	2 × 3	Parotid tumour/pleomorphic adenoma
F	50	Neck	L	0.2	4 × 3	Infected cyst

Table 3. Summary of clinicopathological data of oral and cervical LEC.

	Oral LEC	Cervical LEC
<i>Average age at discovery of lesion (range)</i>	40 (20–68) years	36 (15–61) years
<i>Average duration from the onset of the lesion to consultation (range)</i>	10 (1–36) months	10 (0.2–42) months
<i>Ratio F: M</i>	4:3	7:4
<i>Average size</i>	0.9 cm	4.6 cm
<i>Histopathological features</i>		
<i>Parakeratinized epithelium</i>	07	09
<i>Non keratinized epithelial lining</i>	00	02

**Figure 1.** Clinical view of a palatal lympho-epithelial cyst.

The different presumptive diagnoses proposed by clinicians for oral LECs included mucocoele (2), lipoma (1), fibroma (1), fibro-epithelial lesion (1), and tumor of the accessory salivary glands (1). In contrast, the majority of cervical LECs came with a presumptive diagnosis of branchial cyst (6). However, in two cases the presumptive diagnosis was

neoplasms, such as pleomorphic adenoma and lymphoma. Significantly more often, the clinical diagnosis was correct in the case of cervical LEC, but never with respect to oral LEC ($p = 0.02$). As LECs may clinically and histopathologically mimic neoplasms, awareness among clinicians and pathologists regarding this uncommon entity is important to avoid over-diagnosis and treatment.

The histopathological sections of all oral LECs showed a parakeratinized stratified squamous epithelium, while two cervical LECs were lined by non-keratinized stratified squamous epithelium. In a single oral LEC, the cystic epithelium was seen merging with the surface epithelium. The cystic cavity contained keratin squames and mucinous material, with infected lesions containing neutrophils and macrophages. The pericystic connective tissue contained a dense, chronic inflammatory cell infiltration, predominantly lymphocytic with the presence of one or more germinal centers (Figure 2). None of the cervical LEC with the site of occurrence indicated as the parotid were histopathologically identified to arise within salivary glands and, therefore, the HIV statuses of the patients were not evaluated (Table 2).

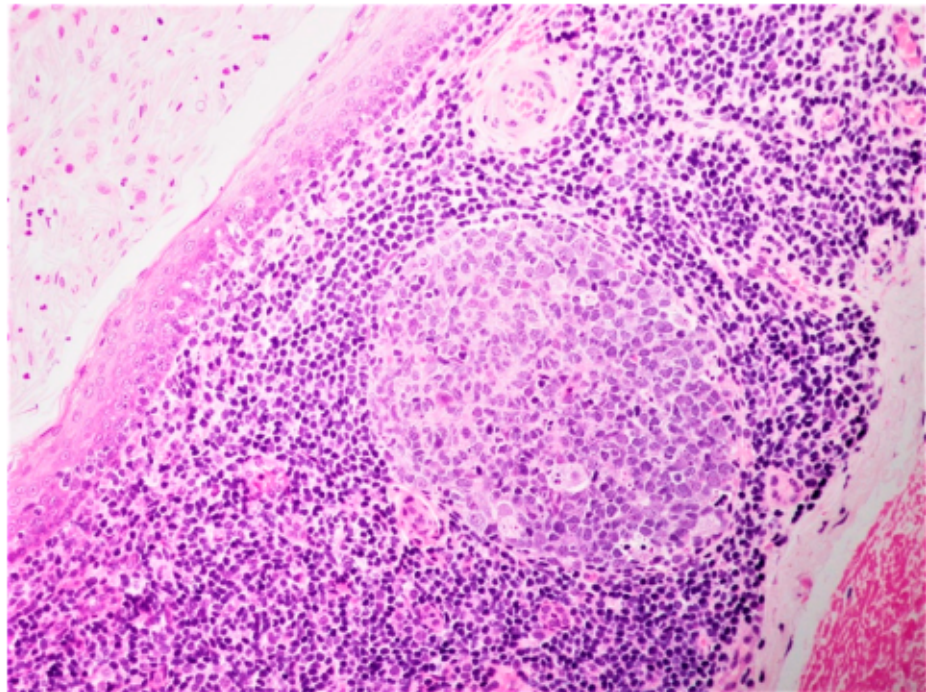


Figure 2. Histological aspect showing epithelial lining, predominantly lymphocytes, with the presence of one germinal center in the cyst wall (H&S $\times 20$).

The surgical resection using a scalpel was performed for each lesion and no recurrences occurred to the best of our knowledge. For cervical LEC, a series of horizontal incisions, known as a stepladder incision, was made to dissect out the lesion. Any preexisting infection was resolved by antibiotics or incision and drainage before surgical treatment was attempted. In the case of oral LEC, complete enucleation of the cyst under local anesthesia was done. According to follow-up information, oral and cervical LECs were indolent lesions that do not in any way contribute to patient survival.

Table 4 [2–4,16–22] presents a summary of case series dealing with more than five LECs. Case reports dealing with single or two lesions amounting to 35 oral LEC are summarized under the Supplementary Materials Table S1 [23–52], while the summary is included in Table 4 last row.

Table 4. Literature-review-based summary of 514 oral LEC.

Reference	Number of Cases	Average Age (Years)	Gender (F:M)	Site	
				T/FOM	Other
2	38	39	15:23	26	12
3	24	36	07:17	24	00
4	120	44	37:83	98	22
16	132	45.5	83:49	112	20
17	77	46.5	54:23	57	20
18	26	33	14:11 *	26	00
19	24	44	09:15	24	00
20	08	40	05:03	07	01
21	21	32	12:09	17	04
22	09	27.6	00:09	09	00
23–51	35	31.68	19:16	21	14
Total/average	514	38.11	255:258	421	93

* Gender for one patient is not given; T/FOM: tongue and floor of the mouth.

A literature review based on 514 cases of oral LEC revealed a mean age of 38 years and a female-to-male ratio of 1:1.1. The majority of oral LEC were found to develop in the tongue and floor of the mouth (Table 4). In addition, similar to oral LEC presented, the literature also indicates that they are small lesions that generally do not exceed more than 2 cm [2–4,16–22].

4. Discussion

Of the seven cases of oral LECs studied, we note an average of 40 years (Tables 1 and 3) compared to 38 years reported according to the literature (Table 4). Cervical LECs were diagnosed at an average age of 36 years, which is slightly above the literature [8,12]. The ratio of female:male for oral LECs is 1.33 (four females to three males), according to our cases (Table 3), and 0.9 (255:258) in the literature (Table 4). Though the majority of studies analyzed reveals female predilection [2,16–18,20,21], one large series of cases from China [4] skewed the general trend, resulting in a slight male predilection. The preferred location of the oral LEC is the tongue and floor of the mouth (Table 4), where three (42%) of our cases occurred, followed by single lesion each at the palate, ventral tongue, and tonsillar fossa. A similar trend was seen in the literature, as well, with respect to oral LEC, with 81% of the total lesions occurring in either the tongue or floor of the mouth (Table 4).

The cervical LECs occur in a great majority of cases in the superior anterior triangle of the neck [15] and parotid region [53]. According to Ford et al. [10], they can be found in all the territory arising from the branchial arches. LECs that arise in the parotid region are unique as they have been shown to be associated with HIV infection and are also described as benign lymphoepithelial lesions (BLEL). However, none of the cases described by Wu et al. [53] were found to harbour HIV or EBV infection. Even so, unlike oral and cervical LEC, similar lesions in the parotid would require investigation to exclude HIV, as the management of such lesions is not purely surgical [54].

Presumptive diagnoses of the seven oral LECs were similar to those in the literature [4]. Considering the rarity of the lesion, none of the oral LECs were clinically diagnosed as such. In addition, bearing in mind the inconspicuous nature of the lesion, an inaccurate clinical diagnosis would not hinder the overall management of oral LEC. Presumptive diagnosis of cervical LEC is similar to that found in the literature as well [13]. Unlike oral LECs, clinicians proposed the diagnosis of branchial cyst for majority of patients. However, considering that two lesions were clinically suspected as neoplasms and one carried the clinical diagnosis of lymphoma, the chance of over-treatment is higher with cervical LECs

compared to oral LECs. Although we do not have data on special investigations that have been used, it is possible that an MRI, a CT-scan, or an ultrasound would refine the diagnosis of presumption. Ultrasound scanning is the first-line method of imaging choice, as the lesions are situated superficially. It will be useful to determine if there are any fistula connecting different organs, such as throat and ear. On the other hand, it is recommended to perform CT or MRI to determine the exact location of the cyst.

The gold-standard treatment for both lesions is surgical excision. No recurrence is report in the literature for oral LECs [4,17]. The risk of recurrence of cervical LECs is 0–22% according to the studies [8,10–12].

The etiopathogenesis of two of the oral LECs described in the present study is compatible with Knapp's hypothesis [1]. Indeed, one of the histological sections presented a cystic epithelium in continuity with the surface epithelium. An obstruction of the canal and a cystic proliferation of the latter can be envisaged.

Histological analysis of cervical LEC is similar to that of oral LEC. Cervical LECs have a stratified squamous epithelium, keratinized or non-keratinized, but may also be related to a respiratory epithelium [7]. The cystic epithelium is separated from the underlying corium by a basal lamina [7]. The connective tissue is the seat of an important inflammatory infiltrate, predominantly lymphocytic, organized in a diffuse band or arranged in germinal centers [18]. However, unlike in oral LEC, histopathological diagnosis is more difficult in cervical LEC because of mimics such as cystic metastasis of oral cancer, or HPV-induced oropharyngeal cancer, thymic cysts, and salivary lympho-epithelial cysts of HIV-positive individuals [55].

Limitations of the study include the fact that no information could be identified to convincingly support the theory of pathogenesis of all oral LEC, and long-term follow-up was not available for most of the cases to determine the outcome. Strengths include the fact that clinicopathological comparison reliably identified some differences between oral and cervical LEC, such as the size of the lesion. In addition, it was also identified that inflamed cervical LEC may mimic malignant lesions and Clinicians should be careful when deciding on definitive treatment based on clinical diagnoses.

Implications: oral LEC were confirmed to be indolent lesions that are generally not related to the HIV status of the patient. Clinicians should be aware that inflamed cervical LEC has a higher chance of getting misdiagnosed as a malignant lesion.

5. Conclusions

Oral and cervical LECs are two histologically similar cysts that occur in two distinct sites. The main difference between the two lesions was identified as the size of the lesion with oral LEC presenting as significantly smaller lesions compared to cervical LEC. The literature review supports the information observed in our study with respect to age of occurrence, site predilections, and size. Cervical LECs, particularly the ones that occur in the parotid region, may require further investigations to exclude BLEL of parotid.

Supplementary Materials: The following supporting information can be downloaded at <https://www.mdpi.com/article/10.3390/app12052525/s1>, Table S1—Literature review based summary of case reports of LEC.

Author Contributions: L.T.: literature review, initial draft of the manuscript; P.R.J.: draft of the manuscript, critical review; U.W.A.M.L.A.: preparation of images; R.B.R.N.M.: critical review of the manuscript; T.L.: conception, critical review of the manuscript. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: The Ethical Committee of the University Hospitals of Geneva, Geneva, Switzerland, approved the protocol (ID 2017-00015), as well as the ERC of the Faculty of Dental Sciences, Sri-Lanka (ERC/FDC/UOP/I/2016/40).

Informed Consent Statement: Patient consent was obtained at the time of taking biopsies (incisional/excisional) for diagnostic purposes so that the material could be used for research following deidentification.

Data Availability Statement: The reported cases can be found at the archives of the Department of Oral Pathology, University of Peradeniya and the Unit of Oral Medicine and Oral & Maxillofacial Pathology, University of Geneva.

Conflicts of Interest: The authors declare no conflict of interest.

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