

Original Article

# Oral and maxillofacial pathologic lesion: retrospective studies on prevalence and sociodemographic features

Nurhayu Ab Rahman\*

Oral Pathology and Oral Medicine Unit, School of Dental Sciences, Universiti Sains Malaysia, 16150 Kubang Kerian, Malaysia.

\* Corresponding author: nurhayu@usm.my

Revised edition: 21/09/2014. Accepted: 21/09/2014. Published online: 17/11/2014.

**Abstract** The aim was to study the prevalence and sociodemographic features of odontogenic, non-odontogenic and salivary glands lesions among patients seen in Hospital Universiti Sains Malaysia. This information is essential to assist clinician in formulating reliable differential diagnosis of such lesion. Data on patient demographics, lesion location, tissue of origin and microscopic diagnosis were extracted from the Laboratory and Diagnosis record registries for biopsy specimen accessioned from year 2000 to 2012. This data was subsequently analyzed based on World Health Organization Classification of Head and Neck Tumours (2005). A total of 748 cases were included in the study. Out of the total number of cases, 367 cases were males and 377 cases were females. Ninety seven cases (13%) were of odontogenic origin, while 90 cases (12%) and 197 cases (26%) were of non-odontogenic and salivary gland origin respectively. Forty five percent of cases involved oral mucosal lesions. The most prevalent odontogenic lesion reported within the twelve years period was radicular cyst and ameloblastoma. Non-odontogenic bone lesion was rarely encountered with it making up less than two percent of total cases reported. Pleomorphic adenoma was the most prevalent benign salivary glands neoplasm reported within similar time period.

**Keywords:** Odontogenic, non-odontogenic, jaw lesion, salivary gland neoplasm, jaw lymphoma, head and neck tumours.

## Introduction

Odontogenic lesions affecting the jaws are frequently encountered in clinical setting among Malaysian population. Nevertheless, there is not much data on prevalence and sociodemographic features of these lesions among Malaysian population. Similar studies have been carried out in Chinese (Jing *et al.*, 2007) and Canadian population (Daley *et al.*, 1994).

Several researches conducted in Malaysian population had focused on malignant lesion affecting head and neck region, in particular oral squamous cell carcinoma. This can be attributed to its consequences of significantly higher morbidity and mortality. In the report by the National Cancer Registry (NCR) for year 2003-2005, mouth cancer in Malaysian population is ranked as the 7th and 3rd

most common cancers for the Indian males and females respectively (Lim *et al.*, 2008).

However, research studies on odontogenic lesion in this part of region are scarce. Till date, PubMed online search on odontogenic lesion in Malaysian population only yields three results (Ngeow *et al.*, 2000; Yeo *et al.*, 2007; Siar *et al.*, 2012). It is important to study these lesions in order to assess geographic differences in incidence of these lesions (Daley *et al.*, 1994) and to assist clinician in making more accurate and reliable clinical judgements in counseling patients before biopsy procedure.

According to a study done in Sri Lanka (Okada *et al.*, 2007), odontogenic tumors, although specific, are relatively uncommon lesions in the oral and maxillofacial region. This is reflected in their findings of 226 cases altogether within a

period of 6 years (1996-2002). The most prevalent tumor was ameloblastoma, consistent with the findings of similar studies in Istanbul, 133 cases in 32 years (Olgac *et al.*, 2006), India, 154 cases in 36 years (Sriram and Shetty, 2008) and Nigeria, 236 cases within 23 years (Ladeinde *et al.*, 2005).

In a study of a Northern Californian population, the most prevalent odontogenic tumor according to their finding was odontomas (826 cases) although this lesion is considered to be hamartomas rather than neoplasm (Buchner *et al.*, 2006). They reported 128 cases of ameloblastoma within the period of 20 years.

The above cited studies mostly extracted data retrospectively and based on specimens sent to single diagnostic institution. In a study of a Queensland population, the data were collected prospectively for the period of 12 months (Johnson *et al.*, 2013). They reported 633 cases of odontogenic lesion with 93 lesions are classified as odontogenic neoplasm. As this study adopts the most recent classification by World Health Organization Classification of Head and Neck Tumours (Barnes *et al.*, 2005), the most prevalent of odontogenic neoplasm is keratocystic odontogenic tumor, followed by ameloblastoma (11 cases within 12 month).

The objective of this study is to retrospectively investigate the prevalence and sociodemographic features of odontogenic lesion, non-odontogenic bone lesion and salivary gland neoplasm that affect the head and neck region based on data of a single university based institution for a period of 12 years duration (2000-2012). This study also acts as an objective assessment of diagnostic workload of Oral Pathology Unit, School of Dental Sciences for future references and strategic management.

## Materials and methods

Data was extracted from the Laboratory and Diagnosis record registries for biopsy specimen accessioned from year 2000 to 2012, at the two main pathology diagnostic laboratories: Department of Pathology, Hospital Universiti Sains Malaysia and Oral

Pathology Unit, School of Dental Sciences, University Sains Malaysia Kubang Kerian Kelantan.

The study only includes specimens from oral cavity consisting of developmental, inflammatory, benign and malignant odontogenic, non-odontogenic and salivary glands lesion affecting the head and neck region, based on the updated WHO classification (Barnes *et al.*, 2005). This study excludes nasopharyngeal carcinoma (NPC) and lesions affecting the thyroid due to their specific characteristics and distribution. Data on patient demographics, lesion location, tissue of origin and microscopic diagnosis were analyzed.

## Results

A total of 748 cases were included in the study. Out of the total number of cases, 367 cases were males and 377 cases were females (in 4 cases, gender not documented). Fifty five percent of cases were classified as inflammatory, followed by malignant and benign neoplasm with 27 and 15 percent respectively (Fig. 1). Ninety seven cases were of odontogenic origin, while 90 cases and 197 cases were of non-odontogenic and salivary gland origin respectively. Majority of the cases, forty five percent, arises from oral mucosa (Fig. 2).

The distribution of odontogenic lesion is as follows: most prevalent odontogenic neoplasm is ameloblastoma with 23 cases including 1 case of recurrent and 2 cases of unicystic ameloblastoma, followed by most common odontogenic cyst which was radicular cyst (20 cases). Other odontogenic cyst includes dentigerous cyst, residual cyst and inflammatory cyst of odontogenic origin (non-specific). Keratocystic odontogenic tumor and other odontogenic lesion were less frequently encountered in this study (Table 1).

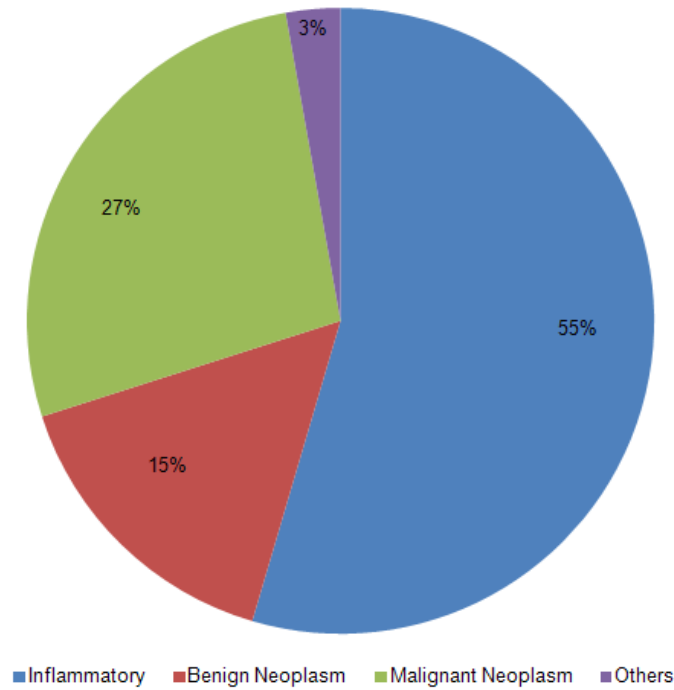
For non-odontogenic lesions arising from the bone only 11 cases encountered within the twelve years period with 7 cases were malignant and 4 cases benign lesion. The distribution as follows, malignant lesion comprised of osteosarcoma (3), chondrosarcoma (3) and Ewing's sarcoma (1), while benign lesion includes cement-

ossifying fibroma and fibrous dysplasia (Table 2). Of interest, 18 cases of different types of lymphoma also reported (Fig. 3).

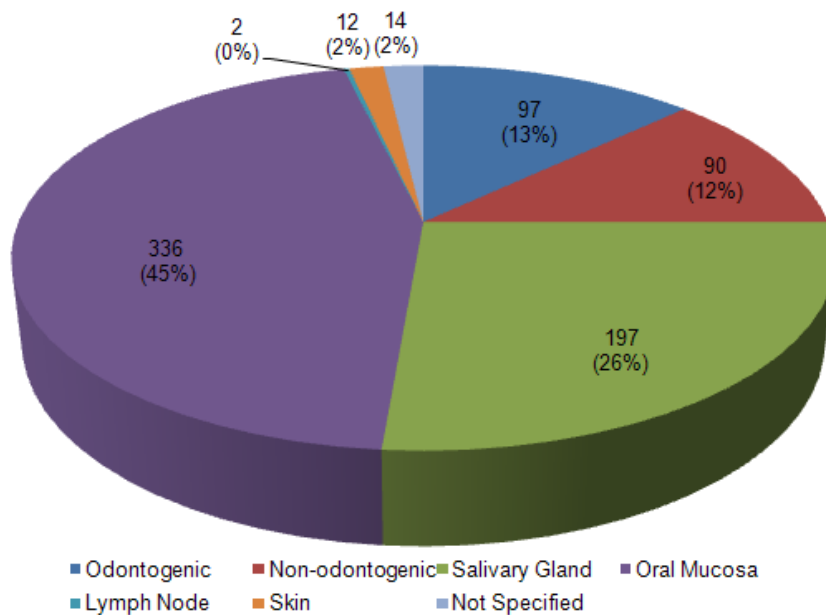
The distribution of salivary gland lesion as follows : out of 197 cases of lesion arising from salivary glands, 109 cases were inflammatory lesion with majority reported as mucous extravasation cyst, 45 cases of benign neoplasm with 39 cases

reported as pleomorphic adenoma and 22 cases of malignant neoplasm (Table 3).

Other findings include 142 cases of malignant neoplasm arising from other structures such as oral mucosa, skin and soft tissue. Majority of these cases reported as squamous cell carcinoma (Table 4) with well-differentiated lesion was most common type reported (Table 5).



**Fig. 1** Distribution of lesion according to aetiology.



**Fig. 2** Distribution of lesion according to tissue of origin

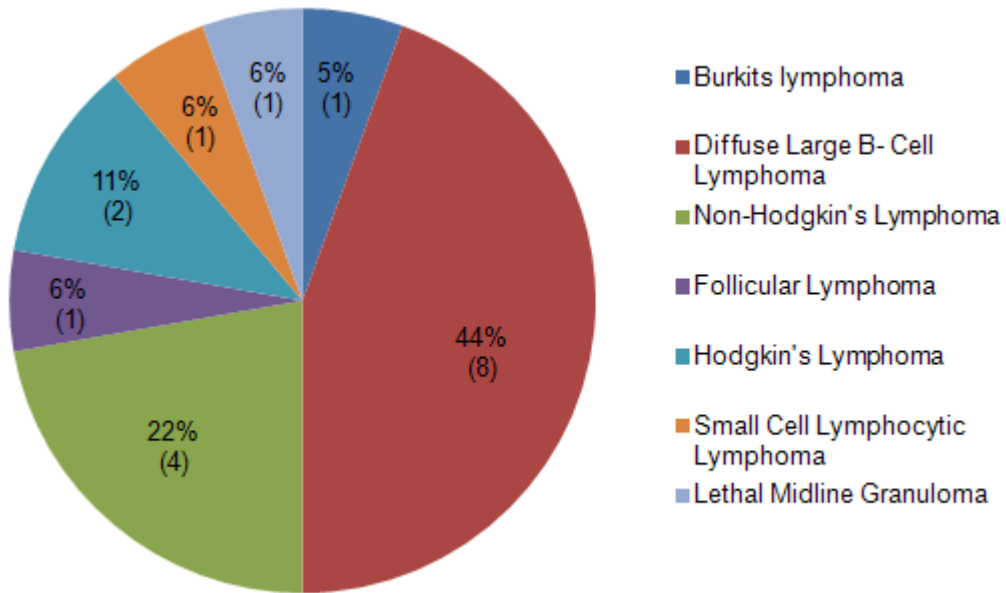
**Table 1** Distribution of odontogenic lesion

Type of odontogenic lesion	No. of cases
Adenomatoid odontogenic tumor	2
Ameloblastic carcinoma	2
Ameloblastic fibrodentinoma	1
Ameloblastoma	23
Amelogenesis imperfecta	2
Cementoblastoma	1
Chronic Hyperplastic Pulpitis	2
Odontome	4
Inflammatory Cyst of Odontogenic Origin	10
Dentigerous Cyst	3
Keratocystic Odontogenic Tumor (Odontogenic Keratocyst)	3
Orthokeratinized Odontogenic Cyst	1
Radicular Cyst	20
Periapical Granuloma	8
Residual Cyst	2
Others*	17
<b>Total</b>	<b>97</b>

Notes - Others\* includes odontogenic lesion such chronic and acute abscess, fibrous scar, chronic granulation tissues etc.

**Table 2** Distribution of non-odontogenic bone lesion

Lesion	Gender	Age	Location	Comments
Osteosarcoma	Female	81	mandible	
	Male	15	not stated	Metastatic
		22	hard palate	
Chondrosarcoma	Female	43	mandible	
		43	mandible	
	Male	21	hard palate	
Ewing's Sarcoma	Male	28	not stated	
Cemento Ossifying Fibroma	Female	13	not stated	
		17	not stated	
Juvenile Ossifying Fibroma		10	mandible	
Fibrous Dysplasia	Male	51	mandible	



**Fig. 3** Distribution of lymphoid lesion.

**Table 3** Distribution of salivary gland lesions

Type of salivary gland lesions	No. of cases	%
Chronic Sialadenitis	13	6.6
Mucocele (Not Specified) / Ranula	35	17.8
Mucous Retention Cyst	6	3.0
Mucous Extravasation Cyst	55	27.9
Pleomorphic Adenoma	39	19.8
Warthin Tumor	6	3.0
Acinic Cell Carcinoma	3	1.5
Adenoid Cystic Carcinoma	8	4.0
Mucoepidermoid Carcinoma	7	3.6
Mucous Secreting Adenocarcinoma	1	0.5
Polymorphous Low Grade Adenocarcinoma	1	0.5
Poorly Differentiated Adenocarcinoma	1	0.5
Recurrent Adenocarcinoma	1	0.5
Others*	21	10.7
<b>Total</b>	<b>197</b>	<b>100</b>

Notes - Other\* - include normal salivary gland tissue, other types of developmental cyst (dermoid cyst, epidermoid cyst, etc)

**Table 4** Different types of other malignant neoplasm reported

Type of malignant soft tissue / mucosa/skin tumors	No. of cases
Leiomyosarcoma	1
Malignant Fibrous Histiocytoma	10
Malignant Fibrous Nerve Sheath Tumor	2
Melanoma	1
Embryonal Rhabdomyosarcoma	2
Squamous Cell Carcinoma	121
Basal Cell Carcinoma	5
Total	142

**Table 5** Details of oral squamous cell carcinoma distribution

Type of Lesions	No. of Cases
Squamous Cell Carcinoma (Not Specified)	11
Well- Differentiated	66
Moderately Differentiated	31
Poorly Differentiated	4
Recurrent	2
Metastatic	5
Basaloid	2
Total	121

## Discussion

The sociodemographic features for solid ameloblastoma is as follows: nineteen cases reported with 13 patients (68%) were female and 6 male patients (31.5%), male to female ratio of 1.8: 1. The minimum age reported was 14 years old female and the maximum age was 65 years old. The mean age at diagnosis was 36.8 years. Fifteen cases arise from mandible (78.9%), one case involved hard palate and maxilla respectively and in two cases, the information of lesion location was not available (Table 6).

Other findings include, one case of recurrent ameloblastoma, which occurred in 31 years old female (lesion arising from mandible) and three cases of unicystic ameloblastoma with the youngest age at diagnosis was 3 years old female (2 cases from mandible and one case reported arising from maxilla). These findings

support the findings of recent study carried out by Siar *et al.* (2012).

Two cases of ameloblastic carcinoma also have been reported. One case reported in 53 years old and 76 years old respectively with both patients are males and lesion arises from mandible (Table 6). The ameloblastic carcinoma is a rare primary odontogenic malignancy that combines the histological features of ameloblastoma and cytological atypia. Fewer than 60 cases have been reported (Barnes *et al.*, 2005).

For central non-odontogenic lesions, eleven cases have been reported. Of particular interest are the malignancies of the jaws. According to Regezi *et al.* (2012), approximately, five percent of osteosarcoma occur in the jaws with the mean age at diagnosis is at 35 years (one to two decades later compared to lesion arising from long bones), with age range from 8 to 85 years old.

**Table 6** Sociodemographic features of ameloblastoma and related lesion

Lesion	Gender	Age	Location	Comments
Unicystic Ameloblastoma	Female	3	Mandible	
		23	Mandible	
		27	Maxilla	
Ameloblastoma	Male	17	Mandible	
		23	Mandible	
		25	Mandible	
		30	Mandible	
			Mandible	
		34	Mandible	
			Not Stated	
		37	Mandible	
		39	Not stated	
		41	Mandible	
		42	Not stated	
		44	Mandible	
		65	Hard palate	
		Female	14	Mandible
	27		Mandible	
	31		Mandible	Recurrent
	47		Mandible	
	50		Mandible	
Ameloblastic Carcinoma	Male	53	Mandible	
		76	Mandible	

\*Ameloblastoma -one case male, age not stated lesion arise from mandible

\*Ameloblastoma- one case male, age and location not stated

**Table 7** Distribution of lymphoid lesion

Lesion	Gender	Age	Location	Comments
Diffuse Large B- Cell Lymphoma	Male	44	Neck	
		78	Sublingual	
		74	Neck	
		58	Not stated	
		19	Neck	
		Female	68	Tonsil
		55	Neck	
B-cell lymphoma	Male	28	Not stated	
Burkitt lymphoma	Male	11	Buccal Mucosa	
Angiolymphoblastic lymphoma	Male	78	Neck	
Follicular lymphoma	Female	42	Neck	
Hodgkin lymphoma	Female	20	Neck	
	Male	55	Neck	
Lethal midline granuloma	Male	45	Hard Palate	
Non-Hodgkins lymphoma	Male	1	Submandibular	
	Female	45	Soft Palate	High Grade
	Female	60	Soft Palate	(Diffuse Mixed Large and Small cell)
Small Cell lymphocytic Lymphoma	Male	6	Soft Palate	

**Table 8a** Sociodemographic features of salivary gland neoplasm

<b>Lesion</b>	<b>Gender</b>	<b>Age</b>	<b>Location</b>	
Pleomorphic adenoma	Female	15	Submandibular	
		16	Parotid	
		22	Parotid	
			Parotid	
			Parotid	
		25	Submandibular	
		26	Parotid	
			Parotid	
		31	Parotid	
			Parotid	
			Parotid	
		34	Parotid	
		40	Parotid	
		44	Parotid	
		46	Parotid	
	49	Submandibular		
	52	Parotid		
	53	Parotid		
	64	Parotid		
	78	Sublingual		
		Not Stated	Parotid	
			Parotid	
		Male	13	Submandibular
			15	Parotid
				Parotid
			22	Submandibular
			24	Submandibular
	33		Submandibular	
			Not Stated	
	34		Parotid	
	36		Parotid	
	42		Not Stated	
	44		Soft Palate	
	48		Parotid	
	50	Parotid		
	72	Submandibular		
		Not Stated	Parotid	
			Parotid	

Note\* One case age and gender not available but arise from parotid



**Table 8b** Sociodemographic features of salivary gland neoplasms

<b>Lesion</b>	<b>Gender</b>	<b>Age</b>	<b>Location</b>	<b>Comments</b>
Adenoid cystic carcinoma	Female	41	Parotid	
		42	Neck	
		54	Submandibular	
		65	Tongue	
	Male	24	Submandibular	
		44	Submandibular	
		68	Upper lip	
Not Stated	68	Not Stated		
Mucoepidermoid carcinoma	Female	22	Parotid	
		34	Hard Palate	
		42	Parotid	
		81	Parotid	
	Male	45	Gingiva	
		72	Parotid	
		73	Not Stated	
Acinic cell carcinoma	Female	61	Parotid	
		Not Stated	Parotid	
		Not Stated	Parotid	
Low-grade polymorphous adenocarcinoma	Female	46	Maxilla	Recurrent
		71	Maxilla	
Basal cell adenocarcinoma	Male	63	Submandibular	
Mucin secreting adenocarcinoma	Male	62	Labial Mucosa	
Poorly differentiated adenocarcinoma	Male	76	Parotid	
Warthin tumour	Male	58	Parotid	
		61	Parotid	
		63	Parotid	
		63	Parotid	
		66	Parotid	
		76	Submandibular	

In our study, three cases (27.3%) have been reported within the 12 year time period, for both osteosarcoma and chondrosarcoma (Table 2). One case of chondroblastic osteosarcoma was reported in 22 years old male arising from hard palate, while 2 cases of metastatic osteosarcoma reported in 15 years old (location of lesion not stated) and 81 year old female arising from mandible. For chondrosarcoma, two cases were reported in 43 year old females, both arising from mandible and one case reported in 21 year old male arising from hard palate

For benign non-odontogenic lesions, such as ossifying fibroma and fibrous dysplasia, these are considered an uncommon lesion in the head and neck region. Cemento-ossifying fibroma tends to occur in the third and fourth decades of life with predilection in tooth-bearing areas of the jaws and slight female predominance (Regezi *et al.*, 2012). In this study, three cases of cement-ossifying fibroma have been reported, all occurred in females (Table 2). For fibrous dysplasia, one case reported in 51 years old male arising from the mandible.

For malignancies arising from lymphoid lesion, eighteen cases have been reported (Table 7). The most common phenotype in the head and neck region was diffuse large B-cell lymphoma with seven cases reported. This lesion occurred in the age range of 19-78 years old with 5 males and 2 female's patient. Three cases were from cervical nodes, others were from sublingual and tonsil areas. Other types of B-cell and T-cell neoplasms also have been reported with age range of 1-78 years old, both involving nodal and extranodal site.

For salivary gland neoplasm, it is considered a rare neoplasm accounting for between 3% and 10 % of head and neck tumours (Jones *et al.*, 2008). According to World Health Organisation, it is classified into 13 benign and 24 malignant neoplasms (Barnes *et al.*, 2005).

In this study, more than half (58%) of these neoplasm reported as pleomorphic adenoma, with the mean age at diagnosis was 33 years old (Table 8a). The age ranges from 13-78 years old with female: male ratio is 1.4:1, slight female

predominance, which is consistent with findings of similar studies in the UK population (Jones *et al.*, 2008). Twenty seven cases (69.2%) arise from parotid glands and 8 cases (20.5%) reported in submandibular gland. One case (2.5%) occurred in minor salivary glands (soft palate) and sublingual gland respectively. Other benign salivary glands neoplasm was six cases (8.9%) of Warthin tumour with five of these cases (83.3%) arising from the parotid glands. The mean age at diagnosis was 65 years old and all cases reported occurred in male patients.

For adenoid cystic carcinoma, eight cases (11.9%) have been reported (Table 8b). Four cases occurred in female, 3 cases occur in male and 1 case, the information on gender is not available. The age ranges between 24-68 years old with the mean age at diagnosis of 51 years old. Three of the cases (37.5%) reported arise from submandibular glands and one case (12.5%) occurred in the parotid. Other site reported includes tongue, neck and upper lip.

Seven cases (10.4%) of mucoepidermoid carcinoma have been reported within the 12 years period with similar gender distribution. The age range was between 22-81 years old with the mean age at diagnosis is 52 years old. Four cases arise from the parotid glands (Table 8b). Both of these findings are also consistent with findings of similar studies in the UK (Jones *et al.*, 2008) and West China (Li *et al.*, 2008) population. Less frequent types of malignant salivary glands neoplasm also have been reported.

## Conclusion

In conclusion, the most prevalent odontogenic lesion reported within the twelve years period was radicular cyst and ameloblastoma. Non-odontogenic bone lesion was rarely encountered with it making up less than two percent of total cases reported. Pleomorphic adenoma was the most prevalent benign salivary gland neoplasm reported within similar time period. There are significant amount of cases involved where the sociodemographic data were not available due to incomplete data registries. This issue

needs to be address accordingly by the relevant authorities as these data is important in prevalence and epidemiological studies of uncommon and rare lesions occurring in the head and neck region.

## Acknowledgements

The author would like to acknowledge Prof. Hasnan Jaafar, former Head of Pathology Department, Universiti Sains Malaysia, Dr. Anani Aila Mat Zin and all staff at Pathology Laboratory, especially Pn. Jamaliah Lin for their help and guidance during the study period. This study was carried out under the Incentive Grant (304/JPNP/600004) provided by Universiti Sains Malaysia.

## References

- Barnes L, Eveson JW, Reichart P, Sidransky D (eds.) (2005). *World Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours*. Lyon: International Agency for Research on Cancer.
- Buchner A, Merrell PW, Carpenter WM (2006). Relative frequency of central odontogenic tumors: a study of 1,088 cases from Northern California and comparison to studies from other parts of the world. *J Oral Maxillofac Surg*, **64**(9): 1343-1352.
- Daley TD, Wysocki GP, Pringle GA (1994). Relative incidence of odontogenic tumors and oral and jaw cysts in a Canadian population. *Oral Surg Oral Med Oral Pathol*, **77**(3): 276-280.
- Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X *et al.* (2007). Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. *Int J Oral Maxillofac Surg*, **36**(1): 20-25.
- Johnson NR, Savage NW, Kazoullis S, Batstone MD (2013). A prospective epidemiological study for odontogenic and non-odontogenic lesions of the maxilla and mandible in Queensland. *Oral Surg Oral Med Oral Pathol Oral Radiol*, **115**(4): 515-522.
- Jones AV, Craig GT, Speight PM, Franklin CD (2008). The range and demographics of salivary gland tumours diagnosed in a UK population. *Oral Oncol*, **44**(4): 407-417.
- Ladeinde AL, Ajayi OF, Ogunlewe MO, Adeyemo WL, Arotiba GT, Bamgbose BO, Akinwande JA (2005). Odontogenic tumors: a review of 319 cases in a Nigerian teaching hospital. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, **99**(2): 191-195.
- Li LJ, Li Y, Wen YM, Liu H, Zhao HW (2008). Clinical analysis of salivary gland tumor cases in West China in past 50 years. *Oral Oncol*, **44**(2): 187-192.
- Lim GCC, Rampal S, Halimah Y (eds.) (2008). *Cancer Incidence in Peninsular Malaysia, 2003-2005*. Kuala Lumpur: National Cancer Registry, Ministry of Health, Malaysia.
- Ngeow WC, Zain RB, Yeo JF, Chai WL (2000). Clinicopathologic study of odontogenic keratocysts in Singapore and Malaysia. *J Oral Sci*, **42**(1): 9-14.
- Okada H, Yamamoto H, Tilakaratne WM (2007). Odontogenic tumors in Sri Lanka: analysis of 226 cases. *J Oral Maxillofac Surg*, **65**(5): 875-882.
- Olgac V, Koseoglu BG, Aksakalli N (2006). Odontogenic tumours in Istanbul: 527 cases. *Br J Oral Maxillofac Surg*, **44**(5): 386-388.
- Regezi JA, Sciubba JJ, Jordan RCK (2012). *Oral Pathology: Clinical Pathologic Correlations*. 6th ed. St Louis: Elsevier Saunders.
- Siar CH, Lau SH, Ng KH (2012). Ameloblastoma of the jaws: a retrospective analysis of 340 cases in a Malaysian population. *J Oral Maxillofac Surg*, **70**(3): 608-615.
- Sriram G, Shetty RP (2008). Odontogenic tumors: a study of 250 cases in an Indian teaching hospital. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, **105**(6): e14-e21.
- Yeo JF, Rosnah BZ, Ti LS, Zhao YY, Ngeow WC (2007). Clinicopathological study of dentigerous cysts in Singapore and Malaysia. *Malays J Pathol*, **29**(1): 41-47.