A pathological retrospective study of 107 cases of non-odontogenic cyst at the Department of Oral & Maxillofacial Surgery, Mahidol University

Yutthasak Kriangcherdsak, Teeranut Chaiyasamut, Natthamet Wongsirichat
Department of Oral & Maxillofacial Surgery, Faculty of Dentistry, Mahidol University

Abstract

Objective: The aim of the study was to analyze the clinical and demographic characteristics of 107 non-odontogenic cysts (NOC) over a 10-year period at the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Mahidol University.

Material and method: All histopathological reports were retrieved from 2003-2012 (10 years). Data were collected and analyzed for age, sex, anatomic location, radiographic and histopathological features.

Results: From the total of 1,978 specimens 107 were NOC, among which the most frequent lesions were mucocele (49.53%), nasopalatine duct cyst (17.76%), and ranula (12.84%). Mucocele was equally present in males and females and was frequently found on the lower lip. Nasopalatine duct cyst and ranula were most common in females with specific locations.

Conclusion: The frequency of NOC in the study population was different from those reported in the literature. The 3 most common NOC were mucocele, nasopalatine duct cyst, and ranula. These cysts accounted for 80.37% of all NOC.

Keywords: non-odontogenic cyst, mucocele, nasopalatine duct cyst, ranula, retrospective study, pathological lesion, pseudocyst, salivary gland cyst

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Corresponding author:
Natthamet Wongsirichat
Department of Oral and Maxillofacial Surgery, Faculty of Dentistry,
Mahidol University,
6 Yothi Street Rachathewee District,
Bangkok 10400, Thailand
Email: natthamet.won@mahidol.ac.th
Tel: 022007777 ext 3333
Mobile phone: +66819095625
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Introduction

Cyst is defined as a pathologic cavity with solid, semisolid, or gaseous contents. Cysts in oral and maxillofacial region can occur in bone and soft tissues. It can be divided into true cyst and pseudocyst by histopathological classification. True cysts are lined by epithelium and surrounded by fibrous connective tissue. Pseudocyst consists of fibrous connective tissue and are not lined by epithelium. In addition, cysts can also be categorized by their origin into odontogenic and non-odontogenic cysts (NOC).

NOC in head and neck region are divided into 4 groups[1]- developmental cyst, pseudocyst of the jaw, cyst of salivary gland, and cyst & pseudocyst of maxillary sinus. (Table 1)

The incidence of NOC is generally low. Demographic profile of NOC has been reported in several countries, including Brazil[2-4], Canada[5], Turkey[6-7], and Kenya[8]. However, there have been no such reports from Thailand. Therefore, the purpose of this study was to study the epidemiology of NOC at the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Mahidol University, during 2003-2012 (10 years).

Materials and methods

This was a retrospective study of all histopathological reports of NOC patients seen and treated at the clinic of the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Mahidol University, from January 2003 to December 2012 (10 years). Ethical approval for this study was permitted by the Head of the Department of Oral and Maxillofacial Surgery. Each case was followed up for age, gender, anatomical location, and histopathological diagnosis. The data were examined for the type of NOC using Excel version 2010 (Microsoft, Redmond, WA).

Results

The results were obtained from the retrospective data of histopathological reports from the patients, who were overlooked at the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Mahidol University, during 2003–2012.9-12 Figure 1 showed the total numbers of head and neck lesions.

Considering the type of NOC following Neville et al classification, Table 2 summarized the data regarding patient's age and sex.

Table 1 Classification or type of non-odontogenic cysts of the oral and maxillofacial region

Developmental cyst	Pseudocyst of the Jaw	Cyst of Salivary Gland	Cyst and pseudocyst of maxillary sinus
1. Palatal cyst of Newborn	1. Simple Bone Cyst	1. Mucocele	1. Antral Pseudocyst
2. Nasolabial Cyst	2. Stafne Bone Cyst	2. Ranula	2. Sinus Mucocele
3. Nasopalatine Duct Cyst	3. Aneurysmal Bone Cyst	3. Mucous Retention Cyst	3. Sinus Retention Cyst
4. Median Palatal Cyst			
5. Follicular cyst of skin (Infundibular cyst)			
6. Epidermoid Cyst			
7. Dermoid Cyst			
8. Teratoid cyst			
9. Thyroglossal Duct Cyst			
10. Branchial Cleft Cyst			
11. Oral Lymphoepithelial Cyst			

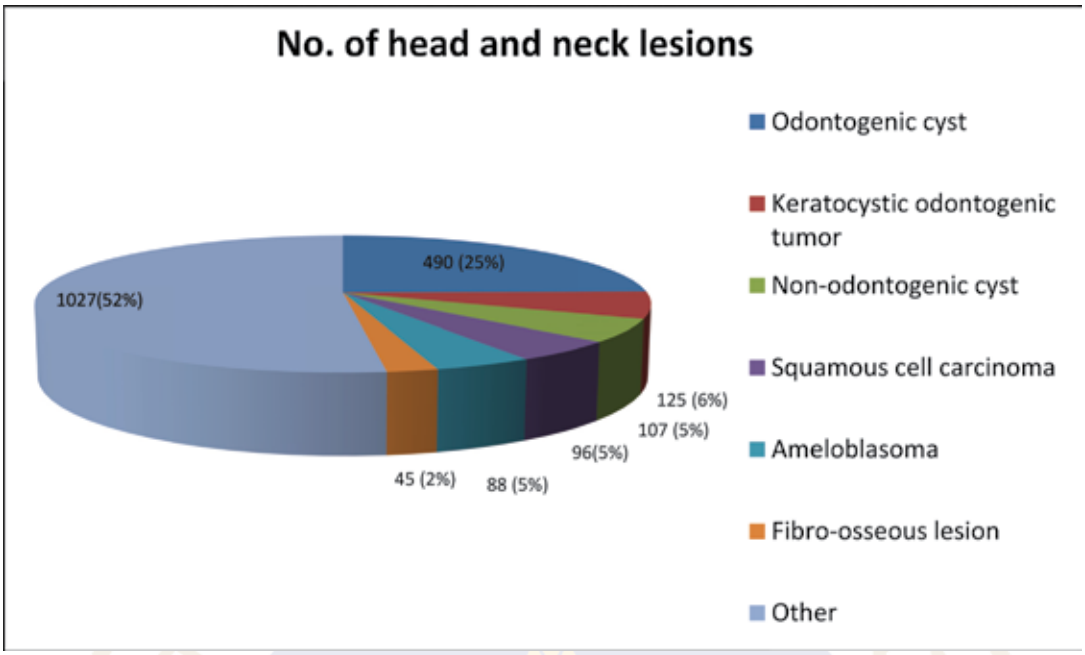

Figure 1 Total number of head and neck lesions observed at the Oral and Maxillofacial surgery clinic

Table 2 Age and sex distribution of patients with NOC

Diagnosis	Case			Percentage	M:F Ratio	Age (years)	
	M	F	Total			Range	Mean
Developmental cyst							
Nasolabial cyst	0	1	1	0.93	-	50	50
Nasopalatine duct cyst	14	5	19	17.76	2.8:1	28-79	52
Infundibular cyst	0	2	2	1.87	-	50-70	60
Dermoid cyst	1	0	1	0.93	-	32	32
Pseudocyst of the jaw							
Simple bone cyst	4	4	8	7.48	1:1	14-79	37
Cyst of salivary gland							
- Mucocele	25	28	53	49.53	1:1.12	8-58	24
- Ranula	1	13	14	12.84	1:13	5-51	25
- Mucous retention cyst	0	2	2	1.87	-	60-61	61
Cyst and pseudocyst of maxillary sinus							
- Antral pseudocyst	3	1	4	3.67	3:1	12-47	34
- Sinus mucocele	0	1	1	0.93	-	29	29
- Sinus retention cyst	1	1	2	1.83	1:1	26-40	33
Total	49	58	107	100	1:1.18	5-79	32

Number

From the total of 107 patients with NOC, there were 49 males (45.79%) and 58 females (54.21%). Figure 2 showed the number of patients based on the type of cyst.

Sex

NOC occurred more in females than males, except nasopalatine duct cyst and antral pseudocyst. However, simple bone cyst, mucocele, and sinus retention cyst were equally found in males and females as shown in Figure 3.

Age

The mean age of the NOC patients was 32 years old, with a range range from 5 - 79 years old. In two patients, the age could not be determined. Patients with simple bone cyst had the widest age range, 14-79 years old. Patients with mucous retention cyst had the oldest mean age, 60.5 years old. The least mean age of 24.15 years was found in mucocele, as shown in Figure 4.

Table 3 showed the age range distribution. Mucocele and ranula were seen with a peak incidence at the age between 10-19 years

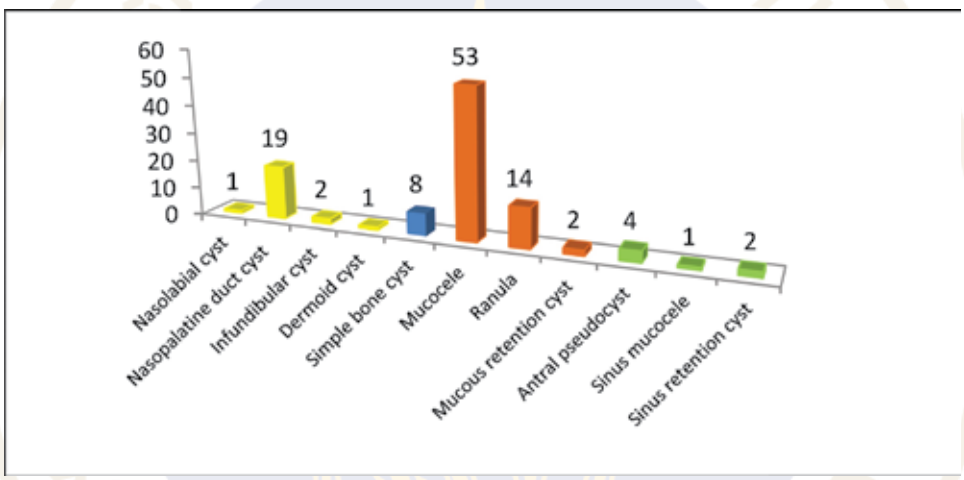

Figure 2 Number of patients based on the type of NOC

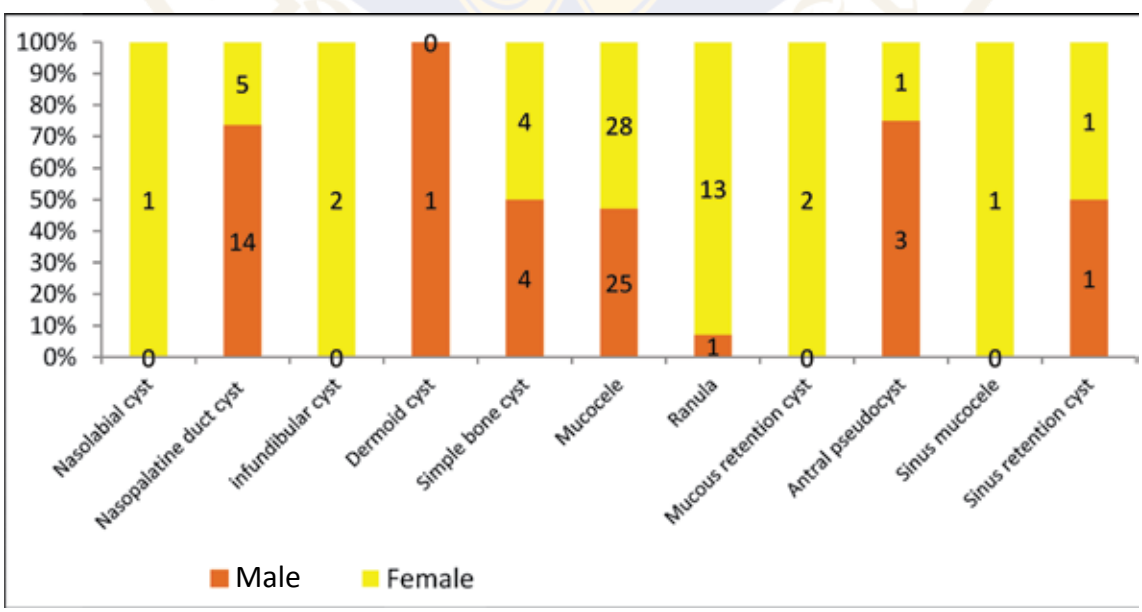

Figure 3 Gender distribution of 107 NOC

(30.19% and 35.71%, respectively). The most common age range of nasopalatine duct cyst were between 30-39 and 70-79 years (21.5%). Simple bone cyst was mostly found in the age range 40-49 years (37.5%).

Location

NOC occurred in the lower jaw more than the upper jaw. The lesions found in the upper jaw were nasolabial cyst, nasopalatine duct cyst, and cyst and pseudocyst of maxillary sinus, while the lesions found in the lower jaw were mucocele, ranula, and simple bone cyst. Mucous retention cyst affected both jaws. There were two patients with infundibular cyst, in one patient it occurred on the right cheek and in other patient it was found at submandibular area. There was only one patient with dermoid

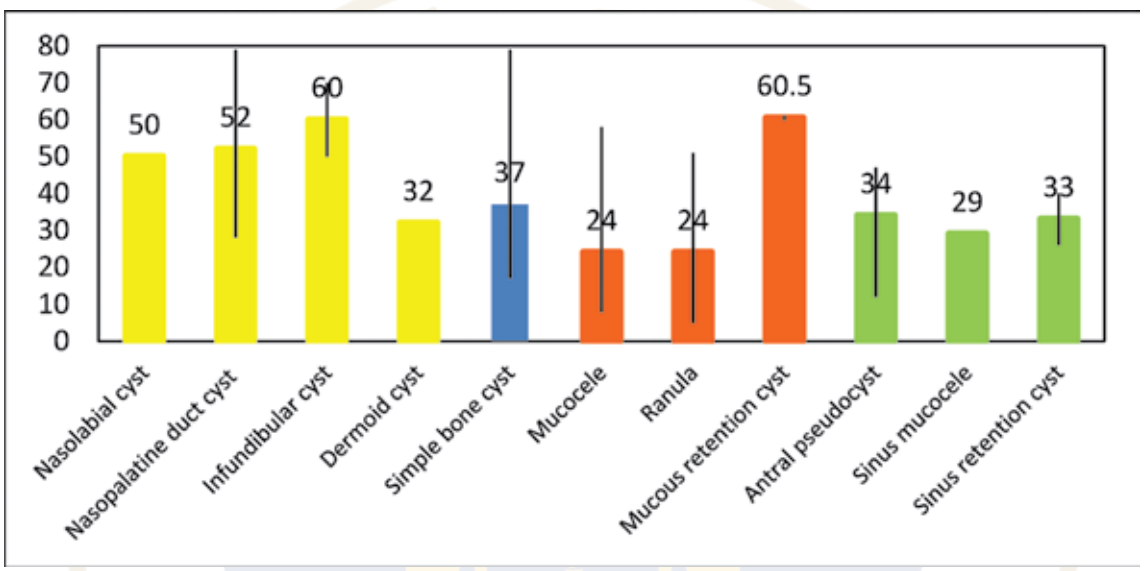

Figure 4 The mean age and age range of 107 NOC patients

Table 3 Age range distribution of NOC

Diagnosis	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70-79	NA
Developmental cyst									
Nasolabial cyst	-	-	-	-	-	1	-	-	-
Nasopalatine duct cyst	-	-	2	4	3	3	3	4	-
Infundibular cyst	-	-	-	-	-	1	-	1	-
Dermoid cyst	-	-	-	1	-	-	-	-	-
Pseudocyst of the jaw									
Simple bone cyst	-	2	2	-	3	-	-	1	-
Cyst of salivary gland									
Mucocele	6	16	15	9	3	3	-	-	1
Ranula	1	5	3	1	1	2	-	-	1
Mucous Retention Cyst	-	-	-	-	-	-	2	-	-
Cyst and pseudocyst of maxillary sinus									
Antral pseudocyst	-	1	-	1	2	-	-	-	-
Sinus mucocele	-	-	1	-	-	-	-	-	-
Sinus Retention Cyst	-	-	1	-	1	-	-	-	-

cyst, which affected the right ear (Table 4).

Discussion

This was a retrospective study regarding the incidence of NOC from histopathological report of patients seen at the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Mahidol University, form January 2003 to December 2012. There were a total of 1,978 specimens attained during the study period, among which there were 107 patients who were diagnosed NOC, with a prevalence of 5.41%. This was much higher than the range of 0.35-1.95%[2-8] reported from other countries.

The 3 most common types of NOC in this study were mucocele (49.53%), nasopalatine duct cyst (17.76%), and ranula (12.84%). These cysts accounted for 80.37% of all NOC. The remaining cysts (19.63%) were nasolabial cyst, infundibular cyst, dermoid cyst, simple bone cyst, mucous retention cyst, antral pseudocyst, sinus mucocele, and sinus retention cyst.

Nasopalatine duct cyst was found in 0.15 - 4% of the oral cysts. In addition, it is the most frequency found developmental cyst (43-100%).[2-8] In this study, 82.61% of patients with developmental cyst had nasopalatine duct cyst. This lesion was more frequently found in males than females, by 2.8 folds, which also corresponded to previous studies[2,4,6-8,13-15]. The average age of patients in this study was 52.47 years, higher than other studies that noted a mean age of 44-48 years.[2,3,7,13-15] In this study, the maximum incidence was between 30-39 years and 70-79 years. Only 19 nasopalatine duct cysts occurred at the anterior maxilla with well-defined radiolucent lesions.

Nasolabial cyst is an oral lesion which occurs very rarely. Patients often present with swelling of the upper lip, resulting in an elevation of the alar of nose. It is commonly asymptomatic.[16] Similarly in this study, a patient presented with a swelling at left alar of nose without any symptoms of pain for 2 months. Sometimes, patients may report of difficulty in breathing and pain.[17-18] Nasolabial cysts have been reported more commonly in females than males by 1.83-2.6 folds.[17,19,22] Most of these cysts occurr between 40-50 years.[17-19,21-22] The only patient in this study was a 50-year-old female. Unilateral nasolabial cysts are usually found, whereas bilateral lesions have been reported in about 10-20%.[18-20,23] This lesion cannot be seen in the radiographic images because it arises in the soft tissue.

Table 4 Localization of NOC according to the site

Diagnosis	No	Location									
		maxilla	mandible	Upper lip/alar	Lower lip	Floor of mouth	Tongue	Buccal mucosa	Maxillary sinus	Palate	Other
Nasolabial cyst	1	-	-	1	-	-	-	-	-	-	-
Nasopalatine duct cyst	19	19	-	-	-	-	-	-	-	-	-
Infundibular cyst	2	-	-	-	-	-	-	-	-	-	2
Dermoid cyst	1	-	-	-	-	-	-	-	-	-	1
Simple bone cyst	8	-	8	-	-	-	-	-	-	-	-
Mucocele	53	-	-	-	42	-	4	5	-	2	-
Ranula	14	-	-	-	-	14	-	-	-	-	-
Mucous Retention Cyst	2	-	-	-	-	-	-	1	-	1	-
Antral pseudocyst	4	-	-	-	-	-	-	-	4	-	-
Sinus mucocele	1	-	-	-	-	-	-	-	1	-	-
Sinus retention cyst	2	-	-	-	-	-	-	-	2	-	-
Total	107	19	8	1	42	14	4	6	7	3	3

Infundibular cyst, also known as **epidermoid cyst**, often arise after localized inflammation of hair follicle. Epidermoid cysts are found in 80% of the skin cyst, and they are most commonly found in the acne-prone areas. Females are affected more frequently than males. These epidermoid cysts should be distinguished from oral epidermoid cysts that occur in the middle region of the floor of mouth and represent a teratoma spectrum. In this study, there were 2 patients with epidermoid cyst, who were aged 50 and 70. The affected areas were the right cheek and left side of chin.

In a retrospective study, Al-Khateeb et al[24] reported 488 patients with cutaneous cyst of the head and neck, in which epidermoid cyst was the most frequently found lesion (49%), followed by pilar cyst (27%), and dermoid cyst (22%). Considering 237 epidermoid cysts, the most frequently affected sites were cheeks (25.74%), neck (24.89%), periorbital area (14.77%), scalp (14.35%), periauricular area(13.08%), and nasal area (7.17%).

Dermoid cyst is a cystic teratoma that contains 3 germ layer derivatives. These teratomas are most commonly found in the ovaries and testes. Two previous studies reported incidence rates of teratomas of the head and neck in the oral cavity as 7% and 1.6-6.5%[25-26]. Al-Khateeb et al[24] reported 109 dermoid cysts, in which the most affected areas were periorbital area (39.45%), neck (19.27%), scalp (14.68%), periauricular area (10.09%), nasal area (8.26%), and cheek (8.26%). Males were accounted more than females. The peak age distribution for dermoid cysts was 0-9 years. In this study, a dermoid cyst was observed in right preauricular area of a 32-year-old male, which had been present for 20 years.

Dermoid cysts that occur at the floor of the mouth are very rare (6.5%). Meyer classified floor-of-mouth cysts into 3 variants by histological criteria- dermoid cyst, epidermoid cyst, and teratoid cyst. Dermoid cysts are lined by epidermis-liked epithelium and the cystic wall contains skin appendage, such as sebaceous gland, hair follicles, or sweat glands. The cystic wall of epidermoid cyst contains no skin appendage. Teratiod cysts are found with skin appendage and other components such as bone, muscle, respiratory, and gastrointestinal tissue[27].

In this study, there were a few patients with epidermoid cyst or dermoid cyst in head and neck region but not in the oral cavity. It is also possible for these cysts to occur in the skin. Patients, who are affected with the cyst in the head and neck region, often see a specialist, such as a dermatologist or ENT doctor, but not a maxillofacial surgeon. Therefore, the incidence of the dermoid cyst can be low.

The etiology of pathogenesis of **simple bone cyst** is unclear. The most widely accepted hypothesis is traumatism. The theory suggests that these lesions develop if intramedullary clots due to trauma do not undergo lysis or resolution[28]. There were 2 cases with a history of trauma and one case had enucleation of a keratocystic odontogenic tumor 6 years ago in the same area as the simple bone cyst. Simple bone cysts are most frequently encountered in first two decades[29], but the findings were different in our study as the cysts were found in the fifth decade. We found no gender preference as reported by Perdigao[30]. Simple bone cysts of the jaw are essentially restricted to the mandible, 6 cases were found in posterior mandible, 1 case in anterior mandible, and another 1 case in both anterior and posterior mandible, but no bone cysts were found in the maxilla. However, there have been reports of the lesion in the maxilla.[31] Mandible has more cortical bone and has a slow repair capability as compared to the maxilla, therefore the lesion is often found in the mandible.

In this study, the incidence of **mucocele** in males was similar to females, which was similar

to several studies that found no differences between male and female[32-34]. Mucocele accounted for 53 cases. They were 25 males and 28 females, with a mean age of 24 years, with the highest incidence in second decade. The results were consistent with many studies that have reported mucocele to occur most frequently in first 30 years, 57-78%.[33] In this study, the most common site was the lower lip 79.25%, buccal mucosa 9.43%, tongue 7.55%, and palate 3.77%. A literature review by Chi et al[33] showed that the most frequently affected sites are lower lip, floor of the mouth, buccal mucosa, ventral tongue, palate, and retromolar area, respectively. It rarely occurs on the upper lip. Mucoceles are most common in children and adult, because young individuals are more likely to experience trauma at the lower lip.

Ranula develops from extravasation of mucous after trauma or obstruction of salivary ducts, and occurs in the floor of mouth. Ranula is predominant in females with a peak prevalence in the 10-29 age group[35-37]; the observations were similar in this study where 92.86% of cases occurred in females and 57.14% of the lesions occur in the patients who were 10-29 years-old. However, there are other studies have shown a male predominance.[36]

Mucous retention cyst is an epithelium-lined cavity that arises from salivary gland tissue. They can occur within the major or minor salivary glands. Intraoral cysts can occur at any minor salivary gland site; they develop most frequently in the floor of mouth, buccal mucosa, lips, and palate.[1,38-40] In this study, there were 2 cases where the minor salivary glands area in the buccal mucosa and palate were affected. Regarding the age of onset, these cysts are common in elderly patients.[38-40] Similarly in this study, the two patients were aged 60 and 61 years. The lesions were asymptomatic but appeared as slow growing swellings.

For the cyst and pseudocyst of the maxillary sinus, there were 4 cases of antral pseudocyst, 1 case of sinus mucocele, and 2 cases of sinus retention cyst. Antral pseudocysts are common and present in 1.5-14 % of the population.[41] In this study, it occurred in 3.67%. They are common seen on panoramic radiographic images, especially in the ceramic panoramic radiographs as the dome-shaped lesion of the sinus floor.[42] Most antral pseudocysts are asymptomatic. Although rare, the affected patients may exhibit pain, paresthesia, or soreness upon palpation. Among the 4 patients with the lesion, 1 patient presented with a painless swelling, 1 patient had periorbital pain, 1 patient had a history of oroantral communication after maxillary molar extraction, but the records of the 4th patient were not found .

The most frequent lesion of the maxillary sinus is the sinus retention cyst.[43] Most retention cysts are small and asymptomatic.[37] Several previous studies have found that patients may have a headache, nasal diacharge, and facial pain.[44-46] We found 2 cases of sinus retention cyst in this study. One patient had no symptoms and the lesion was incidentally found during orthognathic surgery. Another patient had right nasal pain and swelling at the right upper posterior edentulous area for 8 months.

The causes of sinus mucocele are chronic infection, allergic sinonasal disease, trauma, and previous surgery. In some patients, the etiology cannot be identified.[47] Mucocele can slowly expand and destroy surrounding bone. It causes facial swelling, nasal obstruction, and dental pain.[48] In this study, only one patient, who was referred to our department, did not exhibit any symptoms, and the lesion was incidentally found during routine radiographic examination.

If abnormal radiographs are seen, the operator should also consider NOC, besides common lesions such as odontogenic cyst, tumor, and fibro-osseous lesion. The commonly found NOC present with certain characteristic

features. **Mucocele** is common on the lower lip. If there is a swelling on the floor of the mouth, it could be a **ranula**. **Nasopalatine duct cyst** showed well defined radiolucency with round or heart-shaped at anterior palate in radiographs. **Simple bone cyst** most common affect posterior mandible. However, the diagnosis should not only be based on clinical presentation or radiographic finding but also on proper history taking and clinical examination.

As discussed above, the incidence of some lesions in this study differed from other studies. It could have been because of differences in patient demographics and population, and classification of NOC. We, therefore, recommend a similar study in a larger patient population to further evaluate NOC.

In conclusion, there were a total of 107 patients diagnosed with NOC in this study. The 3 most common NOC were **mucocele and ranula, nasopalatine duct cyst**, and **simple bone cyst**, respectively. The patient's age, history of abnormal signs and symptoms, and identification of abnormal radiographic features can help us in diagnosing the lesion and appropriate treatment planning.

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