

# Deep retention mucocele of the retromolar region: a diagnostic challenge with a minimal invasive treatment procedure

**Najwa Karam Genno, DCD, DESS**

Department of Oral and Maxillo-facial Radiology,  
Faculty of Dental Medicine, Lebanese University, Hadath, Lebanon

**Gabriel El-Hajj, DDS, MSD, DESS**

Assistant Professor, Department of Oral Pathology and Diagnosis,  
Faculty of Dental Medicine, Lebanese University, Hadath, Lebanon

## ABSTRACT

Oral mucocele is probably the most common disorder of the minor salivary glands. It is a small-size, benign pathology. It often develops on the lower lip mucosa. In few cases, it grows to an unusual size and is rarely located in the retro molar area, thus hindering the preliminary diagnosis of mucocele. The purpose of this article is to report a case of a large oral mucocele in the retromolar area of a 48-years-old woman and to discuss the differential diagnosis and treatment modalities of such a lesion. The surgical procedure was initially meant to be an incisional biopsy of the lesion; it was modified during the intervention to add a marsupialisation as a means of ultimate treatment.

**Keywords:** case report, marsupialisation, minor salivary gland, oral, retention mucocele, retromolar.

Corresponding author:

Najwa Karam Genno / Email: najwagenno@gmail.com

## INTRODUCTION

Oral mucocele (OM) is the most common benign minor salivary gland lesion.<sup>1</sup> The most common locations of OM are the lower labial mucosa (81.9%), floor of mouth (5.8%), ventral tongue (5.0%), and buccal mucosa (4.8%); infrequent sites include the palate (1.3%) and the retromolar area (0.5%). The lesions are most often described as blue/purple/gray or normal in color. The mean maximum diameter is 0.8 cm (range, 0.1 to 4.0 cm).<sup>2</sup>

Two types of OM can be morphologically distinguished: extravasation mucocele and retention mucocele.

The extravasation mucocele is the most frequent type of mucocele. Its main signs are: predominant location (79%) at the lower lip, age peak in the second decade and more frequent occurrence (in 60%) in the male patients.<sup>3</sup>

The mucous retention cyst of the minor salivary glands, also known as retention mucocele is lined by epithelium. It is probably caused by partial or complete obstruction of the excretory duct. The final formation depends on the amount of the overflowed mucus

and the intensity of the mucus phagocytosis. It usually affects older patients (over 40 years of age), most commonly women and is located in different sites than the extravasation mucocele.<sup>4</sup>

In most cases, diagnosis can be established from clinical details, although a histopathological study is necessary to confirm the diagnosis. The most effective treatment involves complete surgical excision.<sup>5</sup>

This paper reports an atypical case of a large retention mucocele resembling a neoplasm, located in the retro-molar area.

## CASE REPORT

A 48-year-old woman presented to the dental office with a chief complaint of swelling in the posterior right mandible and inability to use her mandibular partial prosthesis. Mild pain of dull character was reported in the region six to seven times during the last four months. It always subsided spontaneously few hours after beginning without painkillers. Four to five episodes of ipsilateral transient paresthesia of the tongue and lower lip were reported in the last month. The swelling had begun six months earlier with a pea-size dimension that has been growing continuously since then. The patient had no history of trauma to the affected area. Her medical records are presented in table 1.

Extra-oral examination showed no asymmetry.

The cervical lymph nodes were not palpable. Upon inspection a 3x2cm tumefaction appeared in

Table 1. Patient's medical records

Medical affection:	Appearance since:	Surgical procedure:	Actual medication:
Goiter	1 year	Thyroidectomy	Levothyroxine, 1 tablet daily
Spinal disk herniation	2 years	Discectomy	
Diabetes mellitus type 2	10 years		Insulin subcutaneous injection for the last 7 years

Table 2. Differential diagnosis according to the clinical situation

	Proposed diagnosis	Support elements	Back away elements
1	Malignant tumor: mucoepidermoid carcinoma/fibrosarcoma	- Firm consistency and adherence to superficial and deep layers - Tongue and lower lip paresthesia - Relatively fast growth - Age and sex	- Transient character of paresthesia - No apparent radiologic involvement - No palpable lymph nodes
2	Benign tumor: peripheral fibroma/neurilemmoma	- Firm consistency - Relatively frequent	- Relatively fast growth
3	Mucocele	- Presence of minor salivary glands - Tender upon palpation and history of mild pain - Mandibular prosthesis wear - Transient character of paresthesia	- No history of trauma - No history of discharge - No fluctuation - No color change - Rare location

the right retromolar area distally to an edentulous alveolar process. The color and surface texture were similar to the surrounding oral mucosa (Fig. 1).

The contralateral region showed normal appearance (Fig. 2).

Upon palpation the tumefaction

felt sensitive. It was non-pulsatile, compressible but not fluctuant; it had a rubbery to firm consistency and did not produce any discharge when manipulated. It also showed a marked adherence to both superficial and deep layers. During the examination, no signs of paresthesia of the tongue



Fig 1. Intraoral 3x2cm soft tissue swelling in right retromolar region. Color and surface texture similar to surrounding oral mucosa.



Fig 2. Contralateral retromolar region showing normal appearance

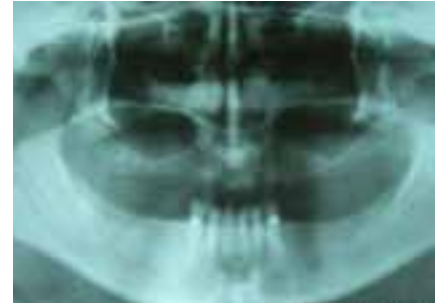


Fig 3. Orthopantomogram showing normal aspect of underlying bone.



Fig 4. Dissection showing the bluish aspect of the roof of the lesion



Fig 5. Translucent yellowish gel emanating from the lumen after perforating the roof of the lesion.



Fig 6. Complete evacuation of intraluminal gel shows a deep cavity and a complete depletion of tumefaction.



Fig 7. Surgical specimen resulting from incisional biopsy.



Fig 8. Wound sutured with gauze in the lumen.



Fig 9. Six months postoperative: patient free from symptoms and clinical signs.

or lower lip could be detected. Furthermore the patient's oral hygiene was satisfactory.

An orthopantomogram showed normal aspect of the underlying mandibular bone (Fig. 3).

Analysis of the clinical situation lead to the differential diagnosis as shown in table 2.

The first differential diagnosis was about malignant lesions; therefore an incisional biopsy under local anesthesia was mandatory. A 1-cm-long incision

was made over the lesion, followed by a gentle dissection to avoid damage to both lingual and buccal nerves. The dissection went four to five mm deep to uncover a smooth bluish membrane (Fig. 4) that, when perforated, gave rise to a translucent yellowish gel (Fig. 5).

The gel was completely evacuated using surgical suction. A 1.5cm deep lumen became obvious (Fig. 6).

An incisional biopsy was then

taken from the tissue constituting the roof of the lumen (Fig. 7) and fixed in 10% formalin.

A gauze was then packed into the lumen and sutured to the mucosal flap in order to marsupialize the lesion (Fig. 8).

Postoperative instructions were given to the patient and the surgical specimen was sent for histopathological evaluation.

Five days later sutures and gauze were removed. The patient reported moderate pain and partial

reduction of the mouth opening capacity. She further went through a series of follow up visits at 15, 30 and 60 days postoperative to control healing process until full epithelium and connective tissue maturity. The last visit was at six months postoperative; the patient was then free from symptoms and clinical signs (Fig. 9).

The histological evaluation of the sections using light microscopy on hematoxylin and eosin stained slices revealed the presence of a regular squamous epithelium and a subjacent fibrous connective tissue including a dilated excretory duct. A minor salivary gland, in the vicinity of the duct, was found. It was surrounded by lymphocytes. The final diagnosis was retention mucocele with no signs of malignancy.

## DISCUSSION

The present case was uncommon in light of the lesion size, which was well above the average reported in the literature, that usually is around one cm in diameter, in addition to the location in the retromolar area.<sup>6</sup>

One of the main interests of this case is the differential diagnosis exercise that it confers. Theoretically, pathologies that may cause swelling in any tissue of the retromolar area should be taken into account. The surface epithelium is obviously intact which discards epithelial pathologies. Moreover based on clinical findings i.e. color and consistency, one would not be disposed to include tumors of vascular or lipomatous origin in the differential

diagnosis.<sup>7</sup> The evolution is relatively fast, six months; this reduces the probability of a chronic process. The evolution, age, sex, consistency, adhesion to both superficial and deep layers, sensory troubles and scarcity of causes of swelling in the region bring us closer to 1- malignancies: primarily the mucoepidermoid carcinoma of minor salivary glands. This pathology is the most common malignant salivary gland neoplasm.<sup>8</sup> The absence of regional lymph node involvement is frequent in this pathology.<sup>7</sup> Other less frequent malignancies could be fibro- or osteosarcoma.<sup>7</sup> 2- benign tumors e.g. fibroma or less frequently neurilemmoma are possible diagnoses especially due to the clinical findings i.e. color and consistency, and high frequency in the case of fibroma<sup>7</sup> or 3- subacute inflammatory process e.g. inflammation due to salivary retention process or pericoronitis on a retained tooth. The pain episodes, tenderness upon palpation and transient paresthesia support this thought but the scarcity of such cases and the mild character of clinical findings back away such diagnoses.

Pericoronitis and osteosarcoma are ruled out due to the absence of radiological findings.

Paresthesia of the right side of lower lip and tongue is in favor of a fast growing process, even though its transient character favors the theory of intermittent, most probably mechanical pressure rather than nerve invasion by a malignant process.

In front of such a large amount of, somehow confusing, information with a malignant tumor in the first row of the differential diagnosis list it was decided to perform an incisional biopsy. The translucent yellowish gel that appeared after perforation of the bluish membrane is that typically seen in OM. The same can also be found in the mucoepidermoid carcinoma as a result of areas of mucus retention.<sup>7</sup> In our case, complete depletion of the tumefaction after suction of the gel almost ruled out the diagnosis of tumor, favoring the OM. So we decided to shift the plan from just an incisional biopsy, for diagnostic purposes, to a treatment modality, the marsupialization that would not jeopardize the nerves and vessels in a region rich with them.

Marsupialisation is conversion of a closed cavity into an open pouch, by incising it and suturing the edges of its wall to the edges of the wound.<sup>9</sup> In this case the closed cavity is the lumen of the OM. It was joined to the oral cavity by a surgically created canal. Interposing the gauze aimed at keeping the canal wide open while epithelial migration, from both cavities, took place to line it. The fluid produced by the minor salivary gland can now freely evacuated to the oral cavity, thus preventing new swelling.

The age, sex, clinical findings i.e. epithelial lining (Fig. 4) and evident lumen (Fig. 6) and histopathological results confirm the diagnosis of retention mucocele. The classification of OM as superficial, classic or deep

has been described according to its distance from oral epithelial lining.<sup>10</sup> In our case the lesion was deep. This explains the firm consistency, the normal overlying mucosal color and the adherence to surrounding tissues. Those same reasons deluded the diagnosis before biopsy taking.

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