

Compendium of Oral Science Volume 10(1)/2023  
Case Report

## Mucocele of lower lip: Case report and literature review

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Received: August 18, 2022  
Reviewed: October 05, 2022  
Accepted for publication: December 01, 2022

### ABSTRACT

*Mucocele is a common pathological lesion in the oral cavity occurring due to obstruction of the salivary gland. The diagnosis is mainly clinical due to its pathognomonic presentation, but it has to be confirmed with histopathological analysis for a definitive diagnosis. A 12-year-old boy went to the Paediatric dental specialist clinic with a chief complaint of a swelling on the left side of the lower lip for about seven months. The aetiological factor was due to trauma secondary to malalignment of an upper canine. The definitive diagnosis was confirmed histologically. The treatment involved surgical resection of the affected tissue, and no recurrence was observed after the surgical procedure. Mucocele is a common soft tissue lesion in the oral cavity and can be successfully resolved via a conventional surgical approach.*

**Keywords:** *Mucocele, swelling, lower lip, excisional biopsy.*

## **BACKGROUND**

The presence of a swelling or lesion in a child's mouth can be rather alarming for both the child and parents. This results in further concern when the lesion appears to be increasing in size. Mucocele is one such lesion that can manifest as a lump anywhere in the oral mucosa (Grover et al, 2020). The term mucocele is derived from the Latin words, mucus and cocele which denotes a cavity containing mucus (Grover, 2020; Yagüe-García et al., 2009).

### **Incidence**

The prevalence of mucocele is 2.5 lesions per 1,000 patients, frequently in the second decade of life (Bentley et al., 2003) and rarely found in children under one year old (Nallasivam et al., 2015). There is no gender predilection for this type of lesion (Grover et al, 2020; de Camargo Moraes et al., 2009).

### **Etiology**

Mucocele is a benign cystic lesion that is caused by a blockage or any damage to the minor salivary glands ducts (Nallasivam & Sudha, 2015). Such blockages can either result from a localized trauma to that region or the presence of salivary duct calculi (Nallasivam & Sudha, 2015). Physical trauma is the main factor that causes the spillage of salivary secretion into surrounding submucosal tissue which later the inflammation may become obvious due to presence of stagnant mucous (Essaket et al., 2020). Persistent oral habits such as lip biting and tongue thrusting are also considered as the aggravating factors that may result in the formation of oral mucocele (Grover, 2020; Nallasivam & Sudha, 2015).

### **Types**

Clinically, there are two types of oral mucoceles which are extravasation and retention type (Essaket et al., 2020). Extravasation type or also known as mucous extravasation cyst occurs due to the leaking of fluid from the salivary gland ducts and acini to surrounding soft tissues as a result of trauma (Essaket et al., 2020). Meanwhile, the retention type occurs due to the obstruction of salivary gland ducts which results in an accumulation of mucous surrounded by the lining epithelium (Essaket et al., 2020). It is frequently seen in major salivary gland ducts. The presence of a mucocele on the floor of the mouth is referred as ranula (Grover, 2020; Nallasivam & Sudha, 2015). The term ranula derived from its appearance which is likened to the underbelly of a frog (Essaket et al., 2020).

### **Clinical features**

Both mucocele and ranula are characterized by a rounded, well-circumscribed, transparent, and pinkish or bluish color lesion of variable sizes (Bentley et al., 2003). The bluish colour of the lesion comes from vascular congestion and cyanosis of the tissue above and the accumulation of fluid underneath (Essaket et al., 2020). The size of the lesion, its proximity to the surface, and the upper tissue elasticity may affect the colour of the mucoceles (Essaket et al., 2020). The lesion may persist from anywhere between a few days up to three years (Valério et al., 2013). Mucoceles are frequently asymptomatic, but sometimes it may cause discomfort as a large lesion can interfere with speech, chewing, or swallowing (Grover., 2020; Hayashida et al., 2010).

Mucoceles can occur at any site of the oral mucosa containing salivary glands (Nallasivam & Sudha, 2015). Extravasation type appears frequently on the lower lip whereas retention type can appear at any other location of the oral cavity and most commonly seen on the floor of the mouth (Grover., 2020; Nallasivam et al., 2015).

The lower lip is the most frequent site for mucocele due to its susceptibility to trauma arising from the upper teeth (Nallasivam & Sudha, 2015). The lesions are also frequently seen on the tongue and buccal mucosa. It is rarely seen in the upper lip, retromolar region, and the posterior third of the dorsum of the tongue (Hayashida et al., 2010). Clinical features and history of the lesion are the most important aspects when trying to diagnose these lesions. History of trauma or oral habits should be explored as these are common aetiological factors for mucoceles (Nagar et al., 2021). Mucoceles also have a tendency to rupture and cause a leak of cystic fluid causing the lesion to shrink in size (Hayashida et al., 2010). Most often, due to the presence of an epithelial lining, recurrence might occur as the lesion gets refilled and increases in size again (Nagar et al., 2021).

### **Histological characteristics**

A histopathologic investigation is important to confirm the diagnosis and to ensure that glandular tissue is completely removed (Hayashida et al., 2010). For retention type, a cyst cavity which is generally well defined with an epithelial wall covered with a row of cuboidal or flat cells produced from the excretory duct of the salivary glands can be found (Bahadure et al., 2012). They show no inflammatory reaction and are true cysts with an epithelial lining (Nagar et al., 2021). Extravasation types are pseudocysts without defined walls. Their mucous is surrounded by a layer of inflammatory cells followed by a reactive granulation tissue made up of fibroblasts caused by an immune reaction. Even though they have no epithelial lining surround the mucosa, they are well encapsulated by the granulation tissues (Bahadure et al., 2012).

### **Treatment**

Surgical excision of the mucocele along with the associated minor salivary gland is carried out when the lesion is symptomatic or causes patient discomfort (Nagar et al., 2021). After appropriate removal, the chances of recurrence are quite low (Grover, 2020). Although surgery is widely preferred, it has several disadvantages such as lip disfigurement and damage to adjacent ducts with further development of satellite lesions (Nagar et al., 2021; Khandelwal & Patil., 2012)

Marsupialization is conducted when the lesion is quite extensive as it prevents a significant amount of tissue loss and also reduces the risk of complications occurring as a result of surgical excision (Grover, 2020). This has certain advantages such as shorter surgical time and shorter post-operative healing period (Bodner et al., 2015). Additionally, patient compliance is also better (Grover, 2020; Khandelwal & Patil., 2012). However, a clinical diagnosis cannot be confirmed through histopathology in this technique (Wu et al., 2011). If it fails, then surgical removal of the lesion is preferred (Bodner et al., 2015).

Micromarsupialization is a technique which consists of draining the accumulated saliva and allowing re-epithelialization of the injured duct along the path of the sutures (Giraddi & Saifi., 2016; Delbem et al., 2000). The purpose of this technique is to reduce the size of the lesion. It is indicated for lesions less than 1 cm in size (Bodner et al., 2015). The micromarsupialization is considered as an ideal treatment in case of the paediatric patient because this technique is simple, rapid, and less chance of recurrence (Giraddi & Saifi., 2016; Delbem et al., 2000). Although this method is simple and non-invasive to vital adjacent structures, recurrence may occur (Nallasivam & Sudha, 2015).

Lasers are also a treatment of choice as it offers advantages of minimizing the trauma, allows rapid tissue healing, and considered as simple mucocele ablation compared with conventional modalities (Besbes et al., 2020). Lasers create a perfect cut with minimum patient discomfort and minimal hemorrhage (Paglia et al., 2015). The operation time is shorter and tolerable by younger patients (Ramkumar et al., 2016). Vaporization with argon and Nd:YAG (neodymium-doped yttrium aluminium garnet) lasers offered a good alternative (Ahad et al, 2017). However, it is difficult to obtain the biopsy sample. Meanwhile, CO<sub>2</sub> laser often results in scarring of the operated site (Besbes et al., 2020). On the other hand, a diode

laser presented with good healing outcomes and it causes no alteration in microanatomy of biopsy specimen (Ahad et al., 2017).

Electrocauterization which is also known as thermal cauterization is refers as a process of generating heat through a resistant metal wire electrode (Besbes et al., 2020). The heated electrode is then applied to living tissue to achieve hemostasis or varying degrees of tissue destruction (Nallasivam & Sudha, 2015). Advantages of electrocautery are minimal bleeding, immediate haemostasis and nearly painless procedure (Bhargava et al., 2014). Disadvantages of electrocautery include need of the anaesthetic agent for cutting, unavoidable burning flesh odour, and low tactile sense (Besbes et al., 2020).

Cryosurgery is a method of lesion destruction by rapid freezing using liquid nitrogen (Bowers & Schaitkin, 2021). This results in the formation of necrotic tissue that allowed to be slough spontaneously, through immunologically mediated mechanisms (Bowers & Schaitkin., 2021). Patients will be able to tolerate this treatment as minimal or no local anaesthesia is used, no bleeding, and minimal to no scarring will be noted after healing (Bowers & Schaitkin, 2021). It has been demonstrated that only two (5.6%) recurrences were observed among 36 mucoceles treated by cryosurgery (Yeh, 2000). There are some disadvantages of postoperative morbidity like edema, irritation, and delayed healing (Bowers & Schaitkin, 2021). Besides that, the operator should also be well trained and competent in handling liquid nitrogen (Bowers & Schaitkin, 2021).

Intralesional steroids have some advantages over the topical ones, including the bypass of the oral mucosal barrier, reducing the chance of mucosal atrophy, and delivery of higher concentrations of drug to the site of the lesion (Sastre & Morges, 2012). The only disadvantage is that intralesional injection is more invasive compared to the topical application of steroids (Sinha et al., 2016).

This case report aims to explain the history, clinical features, and surgical removal of mucocele using a conventional surgical technique.

## **CASE PRESENTATION**

A 12-year-old boy came with a complaint of a big lesion on the left lower lip for the past seven months. Although there was no complaint of pain, the swelling caused some difficulty in eating for the patient. The patient could not recall any trauma to the lower lip. Initially, the swelling was small and gradually increased in size until its present size. The patient and his mother claimed that the lesion never got smaller and neither did it rupture throughout the time that he noticed its presence on the lower lip. The patient had a history of sustained a laceration wound on the scalp due to an alleged fall at age three and he underwent toileting and suturing procedures under general anaesthesia. Otherwise, the patient does not have any other medical problems. The patient had also undergone dental treatment under general anaesthesia without complication when he was five years old.

The extraoral examination was normal. Intraorally, the swelling was seen on the left lower labial mucosa below the vermilion border and extended inferiorly toward the lingual vestibule. Th was about 1.0 cm x 0.8 cm in size, sessile, mobile, pinkish in color, has a smooth surface, and painless which was fluctuant.[Fig 1] The patient also presented with labially displaced upper left permanent canine tooth with a sharp incisal tip.

Based on the history and clinical manifestation, a working diagnosis of mucocele was made. A simple relaxation technique using hypnosis was done to reduce the patient's anxiety. Local anaesthesia was administered (2% mepivacaine with 1:100000 epinephrine (2.2ml)) in the surrounding area of the lesion carefully without penetration of the solution directly into the lesion. Any distortion to the lesion that originated from the anaesthetic solution would make it difficult to obtain a well-defined surgical margin. A suture was placed at the widest part of the lesion and a length of approximately 6 cm of suture was secured using an artery forceps. This holding suture allowed retraction of the lesion during the initial incision which

was followed by blunt dissection of the underlying tissues using scalpel blade no. 15. Multiple nodular-like lesions were seen adhered to the base of the excised tissue. A further undermining of the marginal tissue was done and multiple solitary nodules were removed.[Fig 2] Wound closure was done using resorbable suture size 4.0.

The specimen was submitted for histopathological analysis that confirmed the diagnosis of mucocele. Histopathological examination revealed hyperparakeratinized stratified squamous epithelium with a pool of mucin infiltrated with numerous mucinophages and neutrophils surrounded by thick granulation tissue. [Fig 4 & Fig 5] There are few adjacent lobules of the minor salivary gland exhibiting ductal dilatation and acini atrophy.



**Figure 1: Preoperative view of a dome shaped swelling on the lower lip (1.0 cm x 0.8 cm).**



**Figure 2: Surgical excision of lesion with adhering minor salivary glands.**

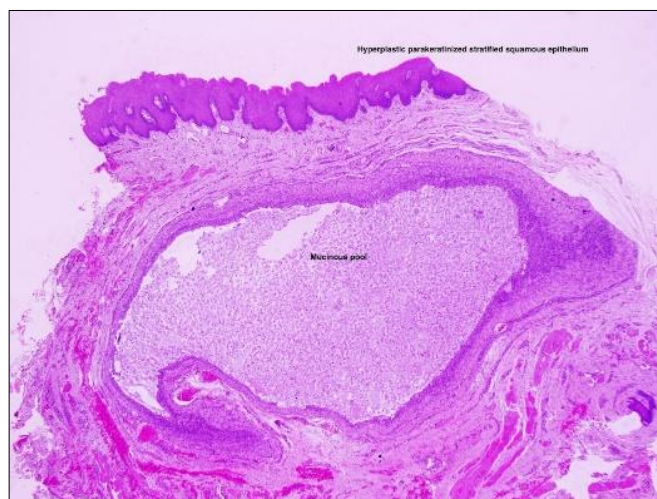
Initially, the patient was reviewed after 1 week for suture removal followed by a review at 1 month and 3 monthly thereafter.[Fig 7] The patient presented with uneventful healing and no recurrence was noted. A soft



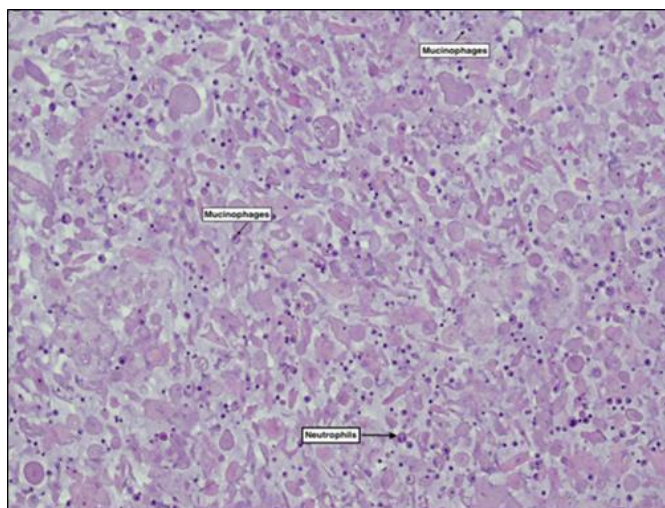
splint was constructed for the patient in lieu of his bruxism. The patient was also referred to Orthodontic specialist clinic for further management of misaligned teeth.



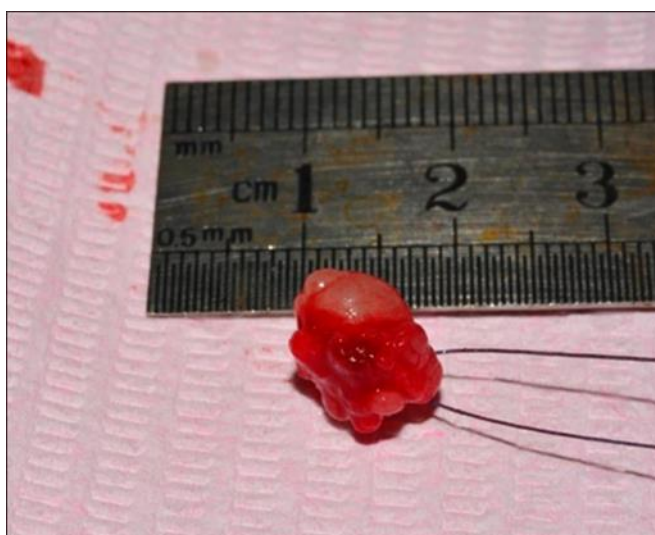
**Figure 3: Post surgical removal of the lesion**



**Figure 4: Photomicrograph showing a mucinous pool surrounded by thick granulation tissue (H&E staining 10x)**



**Figure 5: Photomicrograph showing numerous neutrophils and mucinophages infiltrating the mucin pool (H&E staining 40x)**



**Figure 6: Specimen after surgical removal**



**Figure 7: Post surgical site, 3 months postoperative review.**

## DISCUSSION

Oral mucocele is the seventeenth most frequent oral benign lesion of the oral cavity (More et al., 2014). This lesion can affect all age groups but is frequently seen between the age group of 12 to 20 years (Nallasivam & Sudha, 2015). The two main aetiological factors for mucocele are traumatic injuries and obstruction of the salivary gland duct (Valério et al., 2013). Literature shows that oral habits such as lip biting or sucking, displaced teeth, and severe mechanical trauma is also part of the aggravating factors (Abdel-Aziz et al. 2016; Nallasivam & Sudha, 2015).

In the present case, the patient had a labially displaced upper left permanent canine with a sharp incisal edge that has a high tendency to come into contact with the lip and presumably resulting in chronic stimulation. This was further worsened by the bruxism which patient has been having over the past few years. Thus, it is necessary to remove the aetiological factor by referring the patient to the orthodontic department to correct teeth alignment and constructing a splint while awaiting alignment of teeth (Nallasivam & Sudha, 2015). Apart from that, it was interesting to note that his sister which also under our care had a similar lesion located on the ventral surface of the tongue. That mucocele was successfully removed via a surgical approach.

Oral mucocèles are rarely more than 1.5 cm in diameter and are always superficial (Nallasivam & Sudha, 2015). Mucocele can appear within a few days after minor trauma (Nallasivam & Sudha, 2015). It can either persist unchanged for months unless treated or gradually increase in size (Besbes et al., 2020). The present case showed the continuous persistent presence of mucocèles with a gradually increase in size without rupture for a period of seven months (Besbes et al., 2020).

Histopathologically, mucocèles can be classified into the extravasation type (without an epithelial lining of the cyst wall) and the retention type (with epithelial lining of the cyst wall) (Nallasivam & Sudha, 2015). Mucous extravasation type occurs due to injured salivary gland duct and consequent spillage into the soft tissue around the gland (More et al., 2014). Mucous retention type occurs due to obstruction of salivary ducts commonly because of a sialolith, or periductal scar, or impinging tumor, resulting in the accumulation of saliva in the duct (Besbes et al., 2020). Mucous extravasation type is the most common type of mucocele found in the children, mostly in the lower lip and tongue whereas retention type is rarely seen in children and having common site of occurrence at the palate, the floor of the mouth, and cheek (Besbes et al., 2020).

Lips consist of adipose tissue, blood vessels, connective tissue, salivary glands, and nerves (Besbes et al., 2020). Thus, swelling on the lips can be manifested if there is the presence of any pathology of these tissues (Nagar et al., 2021). However, due to the pathognomonic features of this lesion which can be distinguished based on their clinical appearance, color, consistency, fluctuation, aetiology (history of trauma), and their location of occurrence, the other lesion can be excluded (Nagar et al., 2021). Nevertheless, the excised tissue should always be submitted for pathological investigations to confirm the diagnosis and rule out the salivary gland tumors (Nagar et al., 2021).

Although mucocèles are often painless and do not cause patient discomfort, sometimes, they may interfere with normal oral functions such as speech, chewing, or swallowing and therefore it needs to be removed (Bodner et al., 2015). Likewise, in this case study the mucocele was excised because this lesion interferes with the patient's chewing and eating pattern. The literature describes various treatment modalities for the management of mucocele. Variability in these techniques depends upon the location, accessibility, and size of the lesion (Ayhan et al., 2020). Cryosurgery, intralesional corticosteroid injection, micro-marsupialization, carbon dioxide laser, or conventional surgical removal of mucocele are proposed (Bowers & Schaitkin, 2021; Ahad et al., 2017; Sinha et al., 2016). However, surgical excision is the most common method used to treat mucocele with favorable outcomes (Besbes et al., 2020).

For surgical removal, an elliptical incision is the recommended treatment procedure. This attribute to the fact that this technique will decrease the extent of mucosal tissue loss, minimal fibrous scar formation, and



possibly prevent spilling of the cystic content, which could be responsible for recurrence (Nagar et al., 2021). It is important to avoid injury to the adjacent glands and ducts while placing sutures to avoid reappearance (Abe et al., 2019). Precautions should be taken while suturing the surgical wound to avoid damage to the adjacent glands or ducts with the needle as it may cause recurrence (Bowers & Schaitkin, 2021). This is supported by a study done by Bahadure et al. (2012) which reported that this procedure is the most common followed protocol where in 95.7% of the cases had total remission (Bahadure et al., 2012). Regardless of the chosen technique, complete surgical excision of the lesion together with the surrounding accessory salivary gland is crucial to minimize the risk of recurrence, as well as lesions should be removed down to the muscle layer (Bowers & Schaitkin, 2021).

## **CONCLUSION**

Mucocele is one of the most common benign soft tissue lesions of the oral cavity and it is easily diagnosed based on the clinical appearance and thorough history. Surgical excision with dissection of surrounding and contributing minor salivary gland acini proved to be successful with the least percentage of recurrence.

## **LEARNING POINTS/TAKE HOME MESSAGES**

- Mucocele is one of the most common benign soft tissue lesions of the oral cavity
- Mucocele is easily diagnosed based on the clinical appearance and thorough history.
- Surgical excision with dissection of surrounding and contributing minor salivary gland acini proved to be successful with the least percentage of recurrence.
- The source of the trauma needs to be eliminated to prevent the chance of recurrence.

## **FINANCIAL DISCLOSURE**

Nil.

## **CONFLICT OF INTEREST**

None declared.

## **AUTHORS' CONTRIBUTION**

DFAI, the corresponding author and first author, conceived and provided the data for the case report and submitted the revised manuscript. ZAZ, the second author, collected and organized the data. AV, a co-author, provided logistic support as well as references collections, and WHWAK, a co-author, provided histopathological report and revise the manuscript. All authors have critically reviewed the case report and are responsible for the content and the manuscript.

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