Case Report

Congenital Mucocele of The Lower Lip: Case Report and Literature Review

Gh. Ansari [1], S-H. Daneshvar [2]✉.

[1] Professor, Dental Research Center, Research Institute of Dental Sciences AND Department of Pediatric Dentistry, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran
[2] Assistant Professor, Dental Sciences Research Center, Department of Pediatric Dentistry, School of Dentistry, Guilan University of Medical Sciences, Rasht, Iran

Abstract

Introduction: Mucoceles are described as asymptomatic benign soft tissue masses of the oral cavity. They are soft, smooth, spherical, translucent and fluctuant in the clinical appearance. The most common site of the mucocele is the interior surface of the lower lip followed by the tongue, floor of mouth (ranula), and the buccal mucosa.
Case Reports: This is a case of a 2-year-old girl referred to the Department of Pediatric Dentistry at Shahid Beheshti University of medical sciences with a soft tissue swelling on the lower lip appeared at birth. Typical clinical signs and features along with pathology report lead to the diagnosis of a mucocele. Surgical excision was carried out as the treatment of choice with the histopathological sample being provided for the lab. A spilled mucin surrounded by fibrovascular connective tissue was the lab report. Soft tissue healing of the surgical site was observed with no scaring in a week follow up.
Conclusion: Clinical signs and histopathological report were indicative of a mucocele.
Key Words: Mucocele, Cysts, Mucins, Diagnosis, Salivary glands

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✉ Corresponding author:
S-H. Daneshvar, Assistant Professor, Dental Sciences Research Center, Department of Pediatric Dentistry, School of Dentistry, Guilan University of Medical Sciences, Rasht, Iran

hedyehdaneshvar@gmail.com

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Introduction

Oral mucoceles (OMs) are known as asymptomatic benign soft tissue lesions mostly seen in the oral cavity. Their clinical characteristics include painless, soft, smooth, spherical, translucent, fluctuant single or multiple nodules with minimal irritation [1]. Although they may appear at any age, most OMs are widely seen in the second and third decade of life. Mucoceles have been classified as either extravasation type or retention type. Extravasation type usually occurred when mucin from the ruptured salivary duct (s) is encapsulated in connective tissue around the gland. Retention type is an actual cyst accumulated with mucin and lined by ductal epithelium [2].

Mucocele formation is related to mucus extravasation from accessory salivary glands and usually has a traumatic origin, especially following lip biting [3]. The lower lip is the most common site of the mouth, however, a mucocele can be found in the tongue, floor of the mouth (ranula), and the buccal mucosa [4]. Management of this lesion is through a surgical excision of the surrounding mucosa and glandular tissue down to the muscle layer [5].

There are numerous reports in the literature showing the most frequent location of the mucocele in the mouth (Table1) [2,6-28].

Interestingly the most of the reported cases were in the second and third decades of life and therefore

Table 1. Reported cases of mucocele in 2000-2016 literature

location	Number of cases	Year of publication
Left Lower Lip	8	2001, 2007, 2013, 2014(2)*, 2015(2)*, 2016
Right Lower Lip	4	2012, 2013, 2014(2)*
upper lip	1	2004
Ventral surface of tongue	4	2011, 2012, 2016(2)*
Floor of mouth	1	2014
Buccal mucosa	2	2007, 2014
Right & left lesion in lower lip	1(3§)	2010
Submandibular gland	1	2016
anterior dorsal tongue	1	2014
alveolar ridge	1	2004

***= represent number of articles published in that year; §= represent number of cases.**

collected data from these reports are limited to this age range. Overall, the mucocele occurrence rate was the same in male and female patients.

Case Reports

A 2-year-old girl was referred to the Department of pediatric dentistry at Shahid Beheshti University of Medical Sciences with a chief complaint of unusual swelling in the left area of the lower lip. Reviewing the patient's history revealed that the swelling was present at the birth and gradually grew and attained the current size. There was no report and history of lip bite trauma or any other trauma. The enlarged lesion was clinically located at the inner aspect of the lower lip proximity in tooth#72 region. Oral examination showed oval shaped, solid, fluctuant, palpable lesion with no increase in temperature and normal overlying mucosa. The color of the lesion was pink and approximately 0.4 cm in diameter (Figure 1). The initial differential diagnosis included Oral lymphangioma, Oral haemangioma, and mucocele, however, pathology report was indicative of mucocele.

The treatment plan was to surgically remove of the lesion. The Procedure was explained to the parents and informed consent form was obtained prior to any treatment. Surgical procedure carried out under a local anesthesia using 2% lidocaine with epinephrine 1:100.000; one cartridge (Darupakhsh, IRAN). In order to reduce the chance of reoccurrence and help to control the bleeding, resection was performed from the base of the lesion (Figure 2&3).

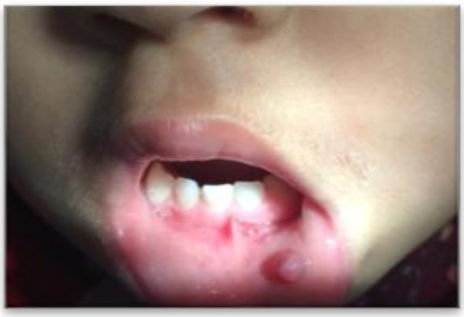

Figure 1. Mucocele in lower lip located in 72 region

Figure 2. The tissue was retained by a tweezers before exclusion

Figure 3. Surgically removed lesion on a gauze

The surgical site was closed using a resorbable suture (Figure 4) and the specimen was sent to the lab in a sealed container with 10% formalin. Postoperative care was instructed including the prescription of analgesics and anti-inflammatories. Follow up visit was arranged a week later at which oral examination revealed normal healing (Figure 5).

Figure 4. Suture placed after the lesion and surrounding tissue was removed

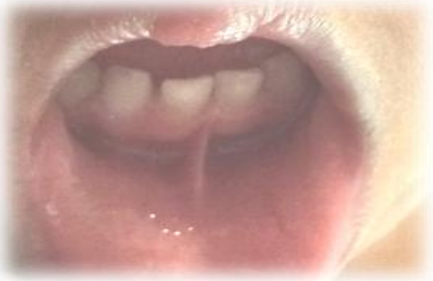

Figure 5. One weak follow up of the surgical site showed relative healing

The histopathological report indicated an area of spilled mucin surrounded by fibrovascular connective tissue, mixed inflammatory cells infiltration mostly foamy histiocytes, hemorrhage, and hemosiderin pigmentation. The lesion was covered by parakeratinized stratified squamous epithelium. All of the signs were indicative of a benign Mucocele.

Discussion

Among numerous types of soft tissue lesions, which could occur in the oral cavity, mucoceles are rare lesions in infants. However, another case of mucocele has been reported in a male neonate [16]. There is no evidence of gender difference in children, adolescents and young adults [2]. Many cases of mucocele have been reported with a wide age range of neonates to adolescents [25]. However, most reported cases were in their second or third decade of life. Trauma and obstruction of salivary gland ducts are the most common etiologic factors in patients suffering mucocele [24]. In neonates trauma to a salivary duct may be attributed to finger sucking in uterus life, traumatic passage through the delivery canal, use of forceps during delivery, or even any manipulation of the neonate after birth [7,29]. Interestingly cases of mucocele in the lower lip are commonly associated with a history of trauma while no history of trauma has been reported when mucocele was found in a rather rare site such as upper lip and buccal vestibule. Orthodontic treatment could be considered as another contributing factor, however, these reports are relatively rare [25]. Tartar-control toothpaste, mouth washing with hydrogen peroxide, deodorant mouthwashes and antiplaque solutions are also known as irritating factors and possible causes of mucocele [30].

Since the secretory duct structure of children are not capable of containing high amounts of secretion, mucous retention cysts are not common in children, whereas mucous extravasation is more frequent [2]. The extravasation mucocele usually occur in younger patients, while mucous retention cysts mostly have been reported in middle aged patients [9]. Despite such a tendency, many diagnosed cases of extravasation type occurred in older patients [14,17,24].

Although the initial diagnosis of the mucocele is based on clinical findings and evident characteristics, computed tomography (CT) and magnetic resonance imaging (MRI) could contribute to more accurate determination of form, diameter, and position of the lesions [30]. Positive mucicarmine staining has also been used to ascertain the presence of extravasated mucin [21]. In a very rare case, a routine ultrasonography showed a male fetus with a cystic lesion measuring 21×11 mm involving the entire oral cavity [20]. MRI showed protruding cystic mass, indicative of a mucocele. Partial aspiration of mucous fluid was performed at 33 weeks of pregnancy. After birth, clinical examination showed presence of a large cystic mass approximately 4 cm in diameter at sublingual region causing protrusion of the tongue. In the current case, there was no prenatal report or indication of mucocele, however, the lesion was reported at birth. Since there was no soreness, irritation, and discomfort, parents had not been seeking any treatment until their daughter was 2 years old.

Any lesion that causes swelling in the lips, can be considered as differential diagnosis. The lip contains adipose, connective tissue, blood vessels, nerves and salivary glands, therefore, pathosis of each of these tissues should be taken into account. Fibroma is a nodular lesion initiated mainly by a trauma and considered as the most common intraoral soft tissue lesion of the lips [18]. Lipomas rarely occurs in oral and maxillofacial regions nevertheless occasionally are mistaken with mucocele [31]. Further differential diagnosis of mucocele includes angiomatous lesions like hemangioma and lymphangioma which can be distinguished using fine needle aspiration biopsy technique [11]. Due to low grade mucin production of mucoepidermoid carcinoma, it may resemble a mucocele on clinical examination [17].

The standard treatment of mucocele is surgical removal of the lesion and there is no difference in the management and curing of retention and extravasation mucoceles [32]. Complications after surgical procedure include fibrous scar formation, temporary paresthesia, and recurrence of the lesion. Fear and lack of cooperation during surgical excision could be an obstacle causing a delay in treatment. General anesthesia may be recommended in these cases, irrespective of the size of the lesion [9]. Cryosurgery has been used for removing mucocele on the floor of the mouth and liquid nitrogen usually used as cryogen agent. Due to its relative lack of discomfort, absence of bleeding and minimal to no scarring, the technique is widely accepted by patients. The main disadvantage of cryosurgery is the lack of a specimen to be examined microscopically for positive confirmation of the diagnosis. Delayed healing, an unpredictable degree of swelling, and lack of precision with depth and area of freezing are other disadvantages of this technique [22]. Micro- marsupialization is usually used in large lesions, however, care should be taken to avoid damage to surrounding structures. In a case report, marsupialization of the congenital mucocele of the ventral face of the tongue was performed at 2 days of age without any recurrence and post-operative complications [20]. One of the major disadvantage of marsupialization is that no further histological examination is possible [32]. Intralesional injection of OK-432, which is a sclerosing agent consist of mixture of a low virulence strain of streptococcus pyogenes incubated with benzyl penicillin, has also been recommended. The use of this technique resulted in the formation of acinar atrophy and consequent healing, however, its side effects include persistent fever, shock and local inflammation [30].

Conclusion

In the present case, all clinical signs along with histopathological report were indicative of a mucocele. The lesion was successfully removed by an excisional surgery and a quick repair was observed at one week follow up.

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