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The role of intraregional dexamethasone injection in the treatment of recurrent tongue mucocele (clinical trial): A case report

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ABSTRACT

Mucocele is the most common minor salivary gland-associated disease of the oral cavity. It involves mucin accumulation causing limited swelling. In recent years, extensive evidence has been published about the usage of intralesional corticosteroid injection in treating mucoceles in adult patients. The aim of the present case report was to assess the effectiveness of intralesional dexamethasone injection in the treatment of mucocele. Two histological types exist: Extravasation and retention. One of the most common oral lesions to be biopsied in pediatric patients is mucocele. Mucoceles can appear at any site of the oral mucosa where minor salivary glands are present. The significant number of cases should be properly conducted in order to check for any prior trauma because the diagnosis is mostly clinical in nature. The lower lip area is where extravasation mucocele most frequently appears, while retention mucoceles can be found at any other site. Mucoceles can affect anybody, however young people are most frequently affected (20-30 years old). Clinically they consist of a soft, bluish and transparent cystic swelling which normally resolves spontaneously. Surgery is commonly used as a kind of treatment. However, other treatments including CO2 laser, cryosurgery, steroid injections and 7 micromarsupialization are also advised. This report reports a case of recurrence of mucocele on the tongue's ventral surface that was unsuccessfully managed five times. Intraregional dexamethasone injection (0.5 ml of 2 g) was found more effective than surgical removal.

Keywords: Dexamethasone, intraregional injection, mucocele, tongue.

1. INTRODUCTION

Oral mucoceles, among the most common oral mucosa lesions, result from trauma to the excretory duct causing the extravasation of saliva (extravasation mucocele) or duct obstruction (retention mucocele) (Ata-Ali et al., 2010). These

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cystic lesions have a dome-like shape and diameters of <1 cm and are typically bluish and fluctuant (De Camargo Moraes et al., 2009). The bluish hue is a consequence of vascular congestion due to the stretching of the overlying tissue; mucoceles may also be pink like normal oral tissue, and the variation in their color depends on factors such as tissue elasticity and proximity to the surface (Baurmash, 2003). Mucoceles occur primarily in the salivary glands of minor origin. About half of mucocele cases start on the lower lip area, as the lip is more prone to trauma; in a study of 312 patients with mucoceles, 73% of the lesions were on the bottom lip, then the tongue (15.4%) (De Camargo Moraes et al., 2009). Mucoceles rarely appear on the upper lip (<5%); when they do, the possibility of neoplasm in the area must be considered (Mustapha and Boucree, 2004).

Despite the fact that some oral mucoceles spontaneously burst, the standard treatment involves surgically removing the lesion and any nearby small salivary glands (Huang et al., 2007); marsupialization can be used for larger lesions and ranulas to avoid the damaging of vital structures (Ata-Ali et al., 2010). Reported mucocele recurrence rates range from 2.8% to 18%, and the removal technique does not affect recurrence (Yamasoba et al., 1990; Oliveira et al., 1993; Yagüe-García et al., 2009; Re Cecconi et al., 2010). Mucoceles on the tongue's ventral surface are most likely to reoccur due to the depth to which associated minor salivary glands are embedded in the lingual musculature and to the possibility of injuring a different minor salivary gland during their removal, causing new mucocele formation (Baurmash, 2003; Choi et al., 2019). In this report, we describe a case of repeated mucocele recurrence on the tongue's ventral surface.

2. CASE REPORT

A 20-year-old middle male presented to the Oral Medicine Clinic at King Saud Dental University Hospital in Riyadh, Saudi Arabia in February 2021, with asymptomatic swelling anterior to the tongue's ventral surface. The swelling had been diagnosed previously as a mucocele and had been treated four times with excisional biopsy but recurred repeatedly. Intraoral examination revealed a pale-bluish firm nodule anterior to the ventral aspect of the tongue. Grossly, the specimen appeared to be a smooth-surfaced, tan-white transparent soft-tissue nodule measuring $0.8 \times 0.7 \times 0.7$ cm. Upon sectioning, thick mucus was observed. Microscopic examination revealed a nodular elevation in the oral mucosa due to the presence of pool of mucin and granulation tissue abutting the overlying atrophic epithelium. The granulation tissue contained neutrophils, muciphages (foamy histiocytes) and reactive myofibroblasts. The surface epithelium was stratified, squamous and of variable thickness and exhibited focal hyperparakeratosis and acanthosis. Skeletal muscle fibers were present at the surgical margin. The diagnosis of mucocele (mucous extravasation phenomenon) was made, although no evidence of salivary gland tissue was seen on the examined sections.

Excisional removal was done five times; thus, the possible feeding gland was not removed and the probability of recurrence was questionable. The patient was then treated by intralesional dexamethasone injection (0.5 ml of 2 g). A follow-up was done after 12 months & then at 18 months the lesion healed completely.



Figure 1 Mucocele on middle ventral tongue surface



Figure 2 During dexamethasone injection



Figure 3 After 1 year follow up showing complete healing.

3. DISCUSSION

Mucoceles of both types are heavily documented in the literature, with a prevalence of 2.5 lesions for every 1,000 people (Yamasoba et al., 1990). Their incidence is highest in decades two and three of life. The diagnosis of mucocele is usually based on clinical findings; lesions such as lipomas and tumors of the minor salivary glands may resemble mucoceles and should be ruled out (Nallasivam and Sudha, 2015). In the current case reported, the diagnosis was based mainly on clinical findings and the patient's previous history of mucocele removal from the area.

Oral mucoceles often recur after surgical treatment and epithelial regeneration due to improper management of the feeding canals. In the case described here, different surgeons had performed a total of five excisions of the patient's original and post-healing recurrent lesions. Two of the pathology reports indicated that the feeding glands not had been identified, which increases the possibility of reoccurrence. There are different treatment methods. However, there are several drawbacks to these surgical procedures, including trauma, pain, deformity of the lip and injury to other critical structures compared with the nonsurgical treatment plan mostly using strong corticosteroids. As observed in a clinical study, full regression was seen in 18 cases out of 20 with oral mucocele using intralesional corticosteroid injections as treatment (Rupam Sinha et al., 2016).

A similar case study was performed, in which seven of the dexamethasone-treated patients were completely healed while either two showed size reduction. Postoperatively, no long-lasting complications were encountered except for one patient's report of local soreness (Baharvand et al., 2014).

4. CONCLUSION

A mucocele is a frequent condition, benign, mucus-filled cyst-like lesion of the minor salivary glands. This report presents a case of recurrence of mucocele on the tongue's ventral surface that was unsuccessfully managed five times. Intraregional dexamethasone injection (0.5 ml of 2 g) was more beneficial than surgical removal. The current study agrees with previous studies. Mucocele recurrence is not well understood, however the site of lesion in ventral surface of the tongue tip that may have exposed the patient to repeated trauma and the multiple recurrences in this patient made this case worthy of documentation.

Informed consent

Written & Oral informed consent was obtained from all individual participants included in the study. Additional informed consent was obtained from all individual participants for whom identifying information is included in this manuscript.

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Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

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