Conservative Surgical Management of Sublingual Ranula: A Case Report and Review of Literature

*Vijay Kumar, 1Anupma Singh, 2Nimmi Singh, 2Archana sudheer

1Department of Oral and Maxillofacial Surgery, R. D. Dental Hospital, India
2Department of Oral Medicine and Radiology, Buddha Institute of Dental Science, Patna

Abstract
Ranula is a rare variant of deep mucoceles of sublingual salivary gland happening on the floor of the mouth with very short history of appearance. It always causes challenging problem for clinician due to their high morbidity and recurrence in previously reported surgical procedures. Combined clinical, radiographic and perioperative evidence of lesion extension can play an important role for selection of definitive surgical procedures. Present study reported clinical presentation, diagnosis, conservative surgical management and outcome of sublingual ranula are discussed.

Keywords
Ranula; Salivary Gland Disease; Conservative Surgical Management of Ranula; Recurrence of Ranula

Introduction
Ranula is a rare variant of mucoceles located on the floor of the mouth and was first described by Hippocrates [1]. It may arise in between gestation to elderly but pick incidence in the second decade [2, 3]. The prevalence of ranula is 2 in 10000 and accounts for 6% of all oral sialocysts related to salivary glands. Out of all cases diagnosed as ranula only 1% to 10% was true retention cysts and rest 90% was pseudo cysts [4, 5]. Clinically there are three types of ranulas namely sublingual, plunging and plunging sublingual [3]. Aspirate of ranula shows high salivary amylase and protein content when compare to serum. This evidence strongly supported origin of ranula from sublingual gland [4]. Principal management for ranula is still via different surgical techniques [6]. As per literature review recurrence rate of ranula with various surgical techniques were as follows: incision and drainage (70% - 100%); marsupialization (36.4% - 89%); limited studies of marsupialization with packing (0% - 25%); excision of the ranula (18.7% - 85%); total excision of the ranula and sublingual gland (0% - 3.8%); and total excision of ranula along with partial sublingual glandectomy was approximately (10%) [6-8]. The aim of present study is to report an additional case of sublingual ranula managed successfully by conservative surgical approach.

Case Description
A healthy 16-year-old girl patient was referred to this centre with the chief complaint of a swelling located on the right floor of the mouth that was interfering with mastication, speech and tongue movement which had 3 weeks previously and has gradually progressed along with intermittent pain. The medical and family histories were unremarkable and there was no history of trauma.

Intraoral examination revealed cystic swelling measuring about 3.5 x 2cm on the right floor of the mouth, below the tongue. It extends from anterior to the posterior on the same side. It was soft in consistency, compressible, fluctuant and tendered on palpation with a bluish oval shaped swelling. On extra oral examination, there was no evidence any palpable neck swelling (Figure 1A).

*Corresponding author: Vijay Kumar, Department of Oral and Maxillofacial Surgery, R. D. Dental Hospital, India. E-mail: vijaypraveenmds@gmail.com Tel No: 9431253479
Received August 2, 2017; Accepted October 5, 2017; Published October 18, 2017


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Non-contrast computed tomography (NCCT) and Contrast-enhanced computed tomography (CECT) of face revealed right aspect of floor of mouth region sharply defined homogenous fluid attenuation cystic solution (4.2 x 1.8 x 2.5 cm) without obvious enhancing margin or enhancing solid mural nodule. The lesion appears lying under the tongue and infero-posteriorly abutting submandibular gland however free from these structures (Figure 1B).

The patient was scheduled for fine needle aspiration cytology (FNAC) and examination of the aspirated fluid revealed high content of salivary amylase and protein and absence of malignant cells. All these findings indicated a benign cystic lesion in the floor of the mouth likely simple ranula.

Total excision of ranula along with partial sublingual glandectomy was performed under general anesthesia. An incision was made over the ranula and 2/3 of cyst was carefully freed from the surrounding tissues by blunt dissection. At that point we felt difficulty in further dissection. Therefore prior to last 1/3 dissection subtotal aspiration was performed then complete dissection was made. Here we noticed capsule of the ranula was firmly attached to the anterior portion of the sublingual gland only and lesion not extended beyond mylohyoid muscle [9]. Therefore just below the capsule attachment a portion of sublingual gland was excised by using diathermy. After separation it was noticed that ranula along with partial sublingual gland was easily separated. Finally incised portion of sublingual gland was sutured by using 3.0 vicryl followed by primary mucosal closer of the same suture. Post-operative recovery was uneventful at 10th days [Figure 1 C] and 6 months [Figure 1 D]. Microscopic examination of excised tissues confirm it as ranula. We have successfully performed the excision of the ranula along with ipsilateral partial sublingual glandectomy with no recurrence, morbidity and good patient tolerance in follow-up.

Discussion

Ranula is a rare variant of deep mucocoeles of salivary gland happening on the floor of the mouth with very short history of appearance [1, 3, 6, 10, 11]. Their incidence is slightly higher in female than in men [10]. Most common clinical features of ranulas are unilateral asymptomatic but gradually enlarging bluish round or oval swelling along with fluctuant and compressible properties. Blue appearance of the ranula may be due to Tyndall effect [1, 3, 6, 10, 11]. Large ranula may causes difficulty in speech, mastication, swallowing, tongue movement, airway obstruction and rarely crossed the midline [3, 6]. It may arise in between gestation to elderly but pick incidence of oral ranula in the second decade and plunging ranula in the third decade [2, 3, 6]. That ranula arises from the sublingual salivary gland is usually large and may extended up to or beyond the mylohyoid muscle but in present case we noticed extension of ranula was up to mylohyoid muscle only [6, 7]. The prevalence of ranula is 2 in 10000 and accounts for 6% of all oral sialocysts related to salivary glands. Out of all cases diagnosed as ranula only 1-10% is true retention cysts and rest ≈ 90% is pseudo cysts [4 - 5]. Most of the features discussed here are similar to present case.

Exact pathogenesis of ranula is still not known but possible pathogenesis of ranula is either due to mucus retention or mucus extravasation of the salivary gland. Former is a true cyst (retention cyst) formation due to ductal obstruction with an epithelial cystic lining and the other is a pseudo cyst formation due to ductal injury and extravasation of mucous without an epithelial lining. In later cases cystic cavity was lined with loose connective tissue and granulation tissue. Apart from rare exception or iatrogenic injury to the submandibular duct, most of the ranulas arise from the sublingual gland [7, 8, 10, 12, 13].

Clinically there are three types of ranulas namely, Simple/Oral/Sublingual ranula: which is the commonest type presenting as an only intraoral component; Plunging/ Cervical/Diving ranula: presents as a cervical swelling crossing beyond the mylohyoid muscle and “tail” sign in computed tomographic is of a clinical importance and mainly located in the right side; and Mixed/Plunging sublingual ranula: which has both sublingual and cervical components [3, 6]. Cystic content of ranula are mainly composed of salivary amylase and protein in higher concentration as compared to serum. Similar finding was also recorded in present case. This finding also supported origin of ranula from sublingual gland rather than submandibular gland [4]. Diagnosis of ranula is largely clinical and in some doubtful cases fine needle aspiration required [5].

Gold standard treatment for ranula is still via complete surgical excision of ranula along with ipsilateral sublingual glandectomy because in this procedure minimal recurrence was recorded [6, 7, 9]. But unfortunately, morbidity rate in this procedure is relatively high due to close proximity of submandibular duct, lingual nerve and...
blood vessels to sublingual salivary gland and at high risk of damage during complete excision of sublingual gland and ranula [8, 9, 11, 14, 15]. Zhao et al in 2005 reported following complications after excision of sublingual gland and ranula; damage of Wharton duct, bleeding, wound dehiscence, postoperative infection and numbness of tongue. According to him overall rate of complication in surgical management of ranula was 1.82%, 0.83%, 0.99%, 0.83% and 4.89% while in case of excision of sublingual gland and ranula it was much higher 2.82%, 1.88%, 1.41%, 1.88% and 11.63% respectively. Here authors also reported overall incidence of recurrence and incidence of recurrence after excision of sublingual gland and ranula only was 5.78% in 450 cases and 1.55% in 129 cases respectively [11].

To minimize this morbidity, need to develop other surgical procedures. During literature review, we got two other surgical procedures namely marsupialization along with gauze packing and excision of ranula along with partial sublingual glandectomy is in light. These procedures having very less morbidity as compare to complete surgical excision of ranula and associated sublingual salivary gland, with accepted recurrence rate. Out of these two procedures rate of recurrence was slightly higher in marsupialization with gauze packing as compare to complete removal of ranula along with partial sublingual glandectomy [5, 7, 9, 11, 15]. Because of its simplicity and complications after excision of sublingual gland, many clinicians prefer marsupialization with gauze packing and complete removal of ranula along with partial sublingual glandectomy as the first treatment option for those cases in which lesion located at or above the mylohyoid muscle [8, 9]. Out of these two procedures we preferred later option because in this procedure rate of recurrence was slightly low. Chung I in 2012 performed ten (9 simple and 1 plunging ranula) partial sublingual glandectomy with excision of ranula and reported only one recurrence particularly in plunging ranula that was treated by total sublingual glandectomy with excision of ranula [8]. Present case was successfully treated by partial sublingual glandectomy with excision of the ranula with no recurrence, morbidity and good patient tolerance in follow-up.

According to recent literature review overall rate of recurrence in incision and drainage, marsupialization, marsupialization with gauze packing, excision of ranula, excision of intraoral or plunging ranula with partial glandectomy and excision of ranula with total sublingual glandectomy was (70% - 100%), (36.4 - 89%), (0% - 25%), (18.7% - 85%), (0% or 100% / 10%) in limited studies and (0% - 3.8%) respectively [6 - 8] and clinical evidence of recurrences appearing between one to twelve months after treatment received [7, 8, 11]. Other nonsurgical treatment option with low rate of recurrence is intracystic injection therapy with OK-432 and Botulin Toxin Type A [7, 11].

**Figure 1.** A. Intraoral photograph of lesion; B. NCCT of lesion; C. 10th days Postoperative photograph and D. 6 months postoperative photograph
Conclusion
The present case report indicated that combined clinical, radiographic and perioperative findings play an important role for selection of different surgical procedures in the management of ranula. In other hand we also noticed no morbidity, no recurrence and good patient tolerance to this procedure in follow-up. Thus we concluded that partial sublingual glandectomy with excision of the ranula is a good conservative treatment option in future for management of the simple / intraoral / sublingual ranula.

References