## Micro-Marsupialization of Sublingual Ranula

# Premna. P. Raj<sup>1</sup>, Rita Zarina. A<sup>2</sup>, Deepa. K<sup>3</sup>

<sup>1</sup>(Junior resident, Dept. of Pedodontics, Government Dental College, Thiruvananthapuram, Kerala, India)
<sup>2</sup>(Professor and Head, Dept. of Pedodontics, Government Dental College, Thiruvananthapuram, Kerala, India)
<sup>3</sup>(Junior resident, Dept. of Pedodontics, Government Dental College, Thiruvananthapuram, Kerala, India)

Corresponding Author: Rita Zarina. A

Abstract: Oral ranula is a retention cyst arising from the sublingual gland on the floor of the mouth as a result of ductal obstruction and fluid retention. Based on clinical features they are of three types; sublingual ranula, plunging ranula and sublingual plunging ranula. The diagnosis may be made based on the clinical presentation, fine-needle aspiration cytology (FNAC), ultrasonography, computed tomography (CT) or MRI. Various techniques used to manage ranulas include incision and drainage, marsupialization, excision of ranula only and excision of ranula along with the involved sublingual salivary gland. Micro-marsupialization is preferred over surgical excision because of the potential surgical complications which may arise from removing the sublingual gland, most notably injury to the lingual nerve, injury to Wharton's duct with the possibility of stenosis leading to obstructive sialadenitis, and ductal lacerations causing salivary leakage. This paper highlights cases of ranulas which have been successfully treated by micro-marsupialization.

**Key words:** Micro-marsupialization, Fine-needle aspiration cytology, Lingual nerve block

\_\_\_\_\_\_

Date of Submission: 04-04-2019 Date of acceptance: 19-04-2019

#### I. Introduction

Ranula refers to a collection of extraglandular and extraductal saliva in the floor of the mouth originating from the sublingual salivary gland (Johan Fagan).

The name "ranula" has been derived from the Latin word "rana" which means "frog". Ranula resembles a frog's translucent underbelly or air sacs. Ranulas are characterized by asymptomatic, well circumscribed, large (>2 cm) cystic cavities and appear as tense fluctuant dome-shaped vesicles, sometimes with a bluish hue. In the floor of the mouth these are typically present unilaterally. Clinically, ranulas are classified as sublingual, plunging and sublingual plunging ranula. Simple ranula is common during the first and second decades of life, while plunging ranula occurs frequently during the third decade of life. Females are more commonly affected than males with a ratio of 1:1.4. <sup>1,2</sup> If left untreated it can cause difficulty in speech and mastication.

The diagnosis of ranula can be done by aspiration technique (FNAC), MRI, ultrasound or CT. In FNAC, the aspirated liquid appears to be highly viscous in nature, with salivary content. A simple ranula can be treated by marsupialization or sclerotherapy or complete excision of the associated salivary gland. Micro-marsupialization is a minimally invasive procedure given by Morton and Bartley for management of ranula, in which the suture is passed from the lesion at its greatest diameter which forms the epithelized tract through which the accumulated saliva gets drained.<sup>3</sup> This article reports two cases of ranulas treated by micro-marsupialization.

#### Case Report-1

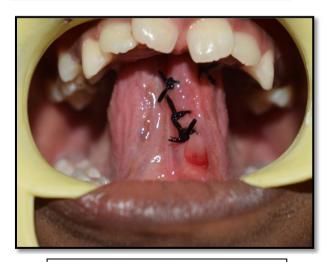
A 9 year old female patient reported to the clinic with a chief complaint of a swelling under the tongue since 2 weeks. There was no history of trauma, infection or known precipitating factors. She also gave a history of discomfort while moving the tongue, speaking and moderate pain due to the swelling, for 1 week. There was no positive history of fever or discharge from the site of swelling. The lesion was initially small which enlarged within a week. Medical history was not relevant. The extraoral clinical examination showed normal features. Intraoral clinical examination revealed a single, large, oval-shaped swelling on the ventral surface of the tongue. The swelling was approximately  $6 \times 3$  cms in size, with capillaries visible on the surface of swelling and had a well defined border, surrounded by normal tissue. On palpation the swelling was non tender, soft in consistency, cystic and fluctuant. There was no evidence of fixity to underlying structure. Based on the history and clinical features, a provisional diagnosis of ranula was made. Aspiration biopsy yielded thick, viscous fluid and histopathological examination (HPE) revealed it to be of mucous nature. The treatment plan focused on solving the main complaint, because the size of the lesion compromised eating and speech, causing severe damage to the physical and social well being of the patient. A conservative approach of micro-marsupialization of the ranula was planned.

DOI: 10.9790/0853-1804126773 www.iosrjournals.org 67 | Page

A routine blood examination was performed before the procedure. Patient was asked to rinse with 0.12%chlorhexidine gluconate solution before the procedure. Lingual nerve block was administered to anesthetize the area. A 2-0 silk suture was passed through the lesion along its widest diameter, taking care not to reach the underlying tissue and a surgical knot was made. Such multiple sutures were placed all over the mucosa which provided vents for the complete draining of the contents. Sutures were then removed after 7 days. The ranula was completely drained, but a smaller swelling recurred after 3 weeks which was again managed similarly. A definite follow up of the patient after the second attempt of micro-marsupialization revealed the ranula to be completely subsided and further follow ups at one month, three months and six months showed a normal picture of the ventral surface of the tongue, and the patient remains asymptomatic.



Pre operative view



Ranula after one week



Reappearance of ranula after three weeks



Management of reappeared ranula



Healing ranula after 4 weeks



Completely healed ranula

### Case Report-2

A 11 year old female patient presented with a complaint of a swelling in the floor of the mouth since 1 week. She also gave the history of discomfort while chewing food, speaking and moderate pain due to the swelling, for 2 days. Medical history was not relevant. The extraoral clinical examination was normal. During the intraoral clinical examination, a single large floating, dome-shaped swelling with bluish hue was observed in the floor of the mouth, extending from 42 to 46 region. The swelling was approximately  $4\times 3$  cms in size, with capillaries visible on the surface of swelling and had a well defined border, surrounded by normal tissue. The swelling was superficial and protruding. On palpation the swelling was mobile, non tender, soft in consistency, cystic and fluctuant. Based on the history and clinical features a presumptive diagnosis of ranula was made. A conservative approach of micro-marsupialization of ranula was planned.

A routine blood examination was performed before the procedure. Patient was asked to rinse with 0.12% chlorhexidine gluconate solution before the procedure. After administering lingual nerve block, the lesion was sutured at multiple points with 2-0 silk. These were removed 2 weeks later, and the lesion was completely resolved. In this patient the ranula healed with the first micro-marsupialization intervention itself and there was no recurrence.





Pre operative views



Sutures placed on ranula



Healing ranula



Post operative view after 3 weeks

#### **II.** Discussion

Ranula was first described by Hippocrates.<sup>4</sup> It is formed either from partial obstruction of a sublingual duct forming an epithelial-lined retention cyst or trauma causing direct damage to the duct or acini, leading to mucus extravasation and pseudocyst formation.

Two schools of thoughts have been described regarding the development of ranulas. One theory states that ranulas develop as a result of mucus extravasation, whereas the second theory is in favor of mucus retention, both as a result of rupture or damage of a duct of sublingual gland. However, current consensus and opinion supports mucus extravasation as the developmental factor because ranulas are mostly devoid of lining epithelium.

Clinically ranulas are of three types. Sublingual ranula Plunging ranula Sublingual plunging ranula

Most common is the "Sublingual ranula", which presents as an intraoral sublingual swelling. The second commonest is the "Plunging ranula", which is located cervically and extends beyond the mylohyoid muscle, and that having both cervical and oral components is known as "Sublingual plunging ranula".

Simple ranulas are more common than plunging types. A simple ranula is formed by localized collection of mucus within the floor of the mouth and may arise from the submandibular duct or from the body of the sublingual gland.<sup>5</sup> Plunging ranulas will attain sufficient fluid pressure to herniate through the mylohyoid muscle and produce a swelling within the neck.<sup>6,7</sup> A plunging (cervical) ranula may be misdiagnosed as thyroglossal duct cysts, vascular malformations, dermoid or epidermoid cysts, and soft-tissue tumors.

The prevalence of ranula is about 0.2 cases per 1000 persons and accounts for 6% of all oral sialocysts. Only 1% to 10% of them are true retention cysts. They usually occurs in children and young adults and the peak frequency occurs in the second decade of life. A study of 83 cases of ranulas in Zimbabwe revealed a high prevalence in HIV positive subjects. Oral ranulas have a higher prevalence in females. According to the literature, the most common site of oral ranula is on the left side of the floor of the mouth. The superficial lesions

0.5 to 1.5 cms in size as compared to ranula arising from the gland with size of >1.5 to 3.0 cms. Typically ranulas are unilateral lesions, and without conservative surgical intervention one cannot determine the offending gland and depth of lesion. They are generally asymptomatic, although large ones can cause aesthetic and functional problems. Very large ranulas can even obliterate the oral cavity, making ventilation and intubation difficult.

The diagnosis of ranula is of clinical importance as some benign and malignant lesions may have similar clinical presentations. There are no specific diagnostic tests for ranulas. Differential diagnosis should be based on the history of the lesion and other investigations.

The differential diagnosis includes cystic hygroma, branchial or thyroglossal duct cysts, laryngocele, various inflammatory and neoplastic lesions of the salivary glands and lymph nodes, granulomatous and adipose tissue diseases, dermoid and epidermoid cysts. <sup>12</sup> The diagnosis may be made based on the clinical presentation, but fine-needle aspiration cytology (FNAC), ultrasonography, computed tomography (CT) and MRI have also been used for differential diagnosis, particularly for plunging ranulas. Contrast enhanced CT scanning shows a characteristic radiolucent 'tail sign' for the plunging ranula, extending from the sublingual gland through the mylohyoid muscle, which differentiates it from other oral ranulas. 13-15 CT and MRI show static images, which cannot assess the active herniation and the nature of the cyst. Jain et al. recommended an ultrasonography for pediatric ranulas, as it does not necessitate sedation or radiation exposure. 16 Ultrasonography is also less costly than CT and MRI imaging. MRI provides more accurate localization of the mass in any plane than ultrasonography for the plunging ranula. FNAC can also provide a definite diagnosis by the presence of yellow aspirate, positive for amylase and mucin. Ranulas originate from the sublingual gland and produces saliva with higher protein concentration as compared to the submandibular gland.<sup>17</sup> FNAC can be performed under local anesthetic in all children.<sup>18</sup> Histopathological examination of the ranula will generally consist of a central cystic space containing mucin and a pseudocyst wall which is composed of loose, vascularized connective tissues. There will be predominance of histiocytes within the pseudocyst wall, but over a period of time these will become less prominent. An important feature in histologic diagnosis is the absence of epithelial tissues in the walls of ranula. 19 A biopsy of the cystic wall is recommended not only for histopathologic diagnosis, but also to rule out the presence of squamous cell carcinoma arising from the cyst wall and papillary cystadenocarcinoma of the sublingual gland, which may present clinically as a ranula. Even though multiple investigative modalities are available, confirmation of the diagnosis via histopathological examination is considered as the gold standard. 20,21

Various treatment modalities advocated are incision and drainage, marsupialization, excision of ranula only and excision of ranula along with the involved sublingual salivary gland. In 1995, Morton and Bartley stated that ranula can be treated by placing silk suture in the dome of the cyst. Later on Delbem et al. utilized the micro-marsupialization technique for the treatment of ranula. This technique involves topical anesthesia of the lesion for 3 minutes and use of a single 4-0 black silk suture passed through the internal part of the lesion along its widest diameter. The suture was removed after 7 days.<sup>22</sup>

Micro-marsupialization is a minimally invasive technique carried out under local anesthesia, the procedure involves draining the accumulated saliva and creating a new epithelized tract along the path of the sutures. The required procedure time is very short, with minimal tissue damage and inflammation. Sandrini et al. performed a modified micro-marsupialization technique for treatment of ranula. The modification consists of an increased number of sutures, decreased distance between the entrance and exit of the needle followed by maintenance of sutures for a longer duration of approximately 30 days. The basic idea of micro-marsupialization is to establish drainage of saliva and formation of new permanent epithelized tract along the path of sutures. They stated the merits like simplicity of execution, low invasiveness and the fact that no special care is required during recovery. This makes the technique a good treatment option, especially in pediatric patients. Haberal et al. suggested that marsupialization of ranula rather than invasive procedures should be the first intervention tried

pediatric patients .<sup>25</sup> Marsupialization, which preserves the sublingual gland and adjacent tissue, is still practiced despite reported recurrence rates as high as 61% to 89%.<sup>25</sup> Micro-marsupialization of oral ranulas by Seton, in which sutures are inserted into the roof of the ranula, was successful in almost all cases.<sup>26–28</sup>The likely explanation for this is that saliva in the ranula leaks away around the sutures, and mucosal epithelium grows along the sutures into the ranula to establish an epithelial-lined tract. This would allow a flow of saliva from the ranula to persist, which would relieve the pressure on the wall, and the granulation tissue in the wall would reduce the size of the lumen until the epithelial-lined tract fuses with the torn glandular end of a duct of Rivinus to create a regenerated duct.<sup>29</sup>

Crysdale et al. reported that the incidence of recurrence after conventional marsupialization of ranulas or pseudocysts of the oral floor was in the range of 61-89%. Therefore they recommended that lesions larger than 1 cm should be treated by excision of the sublingual gland. According to Pandit and Park, about waiting for spontaneous resolution of intraoral ranulas, if the lesion does not resolve, surgical treatment can be recommended. Baurmash advocated that radical surgery should be reserved only for plunging ranula and recurrent cases. 11

CO2 laser and Cr: YSG laser has also been used to vaporize ranulas. Intra cystic injection of sclerotherapy agents like OK-432 (a lypholized mixture of low virulence group streptococcus pyogenes with penicillin G potassium), Bleomycin and Botulinum Toxin Type-A has been reported to be effective in the management of intraoral ranulas. <sup>12</sup>

The reported recurrence rates after various treatment modalities are: incision and drainage (70% to 100%), marsupialization (36.4% to 80%), excision of ranula only (18.7% to 85%), and excision of ranula along with sublingual salivary gland (0% to 3.8%).  $^{25, 33-36}$ 

Though the cases of plunging ranulas have been documented with moderate frequency, failure to differentiate the clinical features of oral and plunging ranulas may be a diagnostic pitfall. These lesions may be difficult to differentiate from benign and malignant salivary gland tumors, especially cystadenocarcinoma and mucoepidermoid carcinoma. So, thorough radiological, biochemical, and histopathological investigations should be carried out for all cases of suspected plunging ranulas.

In these two cases, there were no postoperative complications and have shown that micromarsupialization is a suitable and effective method for management of pediatric intraoral ranulas, whereas in chronic recurrent cases, marsupialization of the ranula combined with total excision of sublingual gland may be preferred.

### **III. Conclusion**

Swelling of the oral cavity can be a diagnostic dilemma for clinicians since a broad variety of lesions that are benign to malignant arise in the oral cavity, especially the floor of the mouth. The diagnosis may be made based on the clinical presentation, fine-needle aspiration cytology (FNAC), ultrasonography, computed tomography (CT) or MRI. Various treatment modalities include incision and drainage, marsupialization, excision of ranula only and excision of ranula along with the involved sublingual salivary gland. Micromarsupialization is a short, simple, less invasive procedure with no special care required during recovery. Hence it can be considered as an effective treatment modality for ranulas in pediatric patients.

### **Bibliography**

- [1]. Morita Y, Sato K, Kawana M, Takahasi S, Ikarashi F. Treatment of ranula-- excision of the sublingual gland versus marsupialization. Auris Nasus Larynx 2003;30:311–14.
- [2]. Yang Y, Hong K. Surgical results of the intraoral approach for plunging ranula. Acta Otolaryngol Stockh 20141342201–05.
- [3]. Rachana P, Singh R, Patil V. Modified micro-marsupialization in pediatric patients: A minimally invasive technique. SRM J Res Dent Sci. 2018;9(2):83.
- [4]. Cedric A. Quick; Seth H. Lowell. Ranula and the Sublingual Salivary Glands. Arch Otolaryngol. 1977; 103(7):397-400.
- [5]. Galloway RH Gross PD, Thompson SH et.al. Pathogenesis and treatment of ranula: report of three cases. J Oral Maxillofac Surg; 1989; 47: 299-302.
- [6]. Davison MJ. Morton RP. Mc Ivor NP. Plunging ranulaClinical observations. Head neck: 1998; 20: 63-68.
- [7]. Mahaddevan M, Vasan N. Management of pediatric plunging ranula. Int J pediatr otolaryngol; 2006; 70: 1049-54.
- [8]. Zhao YF, Jia Y, Chen XM, Zhang WF. Clinical review of 580 ranulas. Oral Surg Oral Pathol Oral Radiol Endod 2004 983 281-7.
- [9]. Chidzonga MM, Mahomva L. Ranula: Experience with 83 cases in Zimbabwe. J Oral Maxillofac Surg 2007; 65(1):79-82.
- [10]. Zhi K, Wen Y, Ren W, Zhang Y. Management of infant ranula. Int J Pediatr Otorhinolaryngol; 2008; 72:823-26.
- [11]. Baurmash HD. A Case Against Sublingual Gland Removal as Primary Treatment of Ranulas. J Oral Maxillofac Surg. 2007 Jan;65(1):117–21.
- [12]. Abdulla Mufeed, Roshni Sajid, Anis Ahmed,, Ashir KR. A fluctuant swelling in the floor of mouth. J Odontol Res 2015;3(1)40-43.
- [13]. Anastassov GE, Haiavy J, Solodnik P, et al. Submandibular gland mucocele: diagnosis and management. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000; 89:159–163.
- [14]. Coit WE, Harnsberger HR, Osborn AG, et al. Ranulas and their mimics: CT evaluation. Radiology 1987; 163:211–216.
- [15]. Charnoff SK, Carter BL. Plunging ranula: CT diagnosis. Radiology 1986; 158:467–468.
- [16]. Jain P, Jain R, Morton RP, Ahmad Z. Plunging ranulas: high-resolution ultrasound for diagnosis and surgical management. Eur Radiol 2010; 20: 1442–1449.
- [17]. Roediger WE, Kay S. Pathogenesis and treatment of plunging ranulas. Surg Gynecol Obstet 1977;144(6):862-4.
- [18]. Zhi K, Wen Y, Zhou H. Management of the pediatric plunging ranula: results of 15 years' clinical experience. Oral Surg Oral Med Oral Pathol Oral Radiol Endodontology. 2009 Apr;107(4):499–502.
- [19]. Batsakis JG, McClatchey KD.: Cervical ranulas. Ann Otol Rh inol Laryngol 1988;97(5 Pt 1):561-2.
- [20]. Engel JD, Ham SD, Cohen DM. Mylohyoid herniation: gross and histologic evaluation with clinical correlation. Oral Surg Oral Med Oral Pathol. 1987;63(1):55-9.
- [21]. Makos C, Noussios G, Peios M, Gougousis S, Chouridis P. Dermoid Cysts of the Floor of the Mouth: Two Case Reports. Case Reports in Medicine. 2011.
- [22]. Verma G. Ranula: A Review of Literature. :6.
- [23]. Sagari S, Shah D, Singh V, Saawarn S, Vamsi K, Patil G. Micro-marsupialization: A minimally invasive technique for mucocele in children and adolescents. J Indian Soc Pedod Prev Dent. 2012;30(3):188.
- [24]. Sandrini FA, Sant'ana-Filho M, Rados PV. Ranula manage ment: Suggested modifications in the Micro-marsupialization technique. J Oral Maxillofac Surg 2007;65(7):1436-8.
- [25]. Haberal, I., Göçmen, H. & Samim, E. (2004) Surgical management of pediatric ranula. Int. J. Pediatr. Otorhinolaryngol., 68, 161-163.
- [26]. Morton RP, Bartley JR. Simple sublingual ranulas: pathogenesis and management. J Otolaryngol 1995;24:253–254.
- [27]. Delbem AC, Cunha RF, Vieira AE, Ribeiro LL. Treatment of mucus retention phenomena in children by the micro-marsupialization technique: case reports. Pediatr Dent 2000;22:155–158.
- [28]. Hoggins GS, Hutton JB. Congenital sublingual cystic swellings due to imperforate salivary ducts. Two case reports. Oral Surg Oral Med Oral Pathol 1974;37:370–373.
- [29]. Harrison JD. Modern management and pathophysiology of ranula: literature review. Head Neck 2010;32:1310-1320.
- [30]. Crysdale, W.S., Mendelsohn, J.D. & Conley, S. (1988) Ranulas mucocoeles of the oral cavity: experience in 26 children. Laryngoscope, 98, 296-298.
- [31]. Pandit, R.T. & Park, A.H. (2002) Management of pediatric ranula. Otolaryngol. Head Neck Surg., 127, 115-118.
- [32]. Yoshimura Y, Obara S, Kondoh T, Naitho SI, Scow SR. A comparision of three methods used for treatment of ranula. J Oral Maxillofac Surg 1995;53(3):280-3.
- [33]. Zhao YF, Jia J, Jia Y. Complications associated with surgical management of ranulas. J Oral Maxillofac Surg 2005; 63 (1):51-4.
- [34]. Parekh D, Stewart M, Joseph C, Lawson HH. Plunging ranu la: a report of 3 cases and a review of liteature. Br J Surg 19 87;74(4):307-9.
- [35]. Dhaif G, Ahmed Y, Ramaraj R. Ranula and the sublingual sali vary glands: Review of 32 cases. Bahrain Med Bull 1998; 20(1):3-4.

Premna. P. Raj. "Micro-Marsupialization of Sublingual Ranula." IOSR Journal of Dental and Medical Sciences (IOSR-JDMS), vol. 18, no. 04, 2019, pp 67-73.