The anatomical basis and rational for the transoral approach during the surgical excision of the sublingual salivary gland for the management of plunging ranula

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Abstract

Purpose: The aim of this study was to illustrate and confirm that the complete intraoral excision of the sublingual gland alone, is anatomically the most rational approach, for the management of plunging ranula.

Material and methods: We retrospectively reviewed clinical charts of diagnosed HIV-infected patients presenting with plunging ranula. The intraoral approach was used for the complete excision of the sublingual gland with evacuation of the pseudocystic content only. Neither extra oral approach, nor ranula dissection / drainage was performed. The surgical procedure was performed using local anesthesia.

Pre- and postoperative MRI-scan investigations were recorded. Histological reports were documented to confirm the diagnosis of oral mucocele. Patients were clinically monitored.

Results: We identified 90 adults presenting with oral mucocele, type ranula. Seventy (77%) of them were diagnosed with HIV infection. Plunging ranula was recorded in 35 (50%) patients from the latter group. The study enrolled 11 operated patients whose files contained useable data, including an acceptable follow-up period. The postoperative follow-up period ranged from three to 15 months. The clinical and postoperative MRI-scans of operated patients demonstrated satisfactory results. There were neither postoperative complications nor recurrence of ranula reported.

Conclusion: The location of the sublingual gland in the floor of the mouth coupled with the physio-pathogenesis of the plunging ranula, makes the transoral complete excision of the offending gland, with the intraoral evacuation of the pseudocyst, anatomically the most rational approach for plunging ranula management. There is no need for cervical approach, ranula dissection and/or postoperative placement of drainage.

Keywords: Plunging ranula; HIV infection; Transoral surgery

1. Introduction

Plunging ranula (PR) is a clinical description of an oral mucocele that arises from the floor of the mouth. In Contrast to the simple or superficial ranula that remains limited to the supra mylohyoid muscle spaces, the PR variety extends below and/or beyond the infra mylohyoid muscle spaces (e.g., the neck).

The commonly accepted physio-pathogenesis of the oral mucocele has been, until recently, mainly traumatic and/or obstruction of the salivary gland excretory apparatus. However, oral mucoceles, and especially the ranula type, have been also reported in the literature in association with Human Immunodeficiency Virus (HIV) infection [[1], [2], [3], [4]]. The exact physio-pathogenic mechanism of this association is, however, unclear. Nevertheless, the high number of PRs observed in association with HIV infection remains of great clinical interest [1,2,4].

Various treatment modalities have been described in the literature. The transoral complete surgical excision of the ipsilateral sublingual salivary gland (SLSG), with only the evacuation of the pseudocystic content, appears to be the most recommended treatment protocol [[5], [6], [7], [8], [9], [10], [11], [12], [13], [14], [15], [16]]. This approach is, on the one hand, based on the commonly accepted underlying physio-pathogenic theory of the origin of the oral mucocele from the SLSG. The approach is also based, on the other hand, on the anatomical location of the SLSG in relation to other anatomical structures in the floor of the mouth. Indeed, the SLSG lies in the submucosal plane, just covered by a thin layer of oral mucosa [17].

However, despite the growing body of evidence on the validity of the intraoral excision of the ipsilateral SLSG with only the evacuation of the ranula during the management of PR, there are decades later, literature reports that continue to advocate for the use of transcervical approach with dissection and/or excision of the ranula pseudocyst [8,[18], [19], [20], [21], [22]].

The aim of this study is to provide clinical evidence-based arguments that illustrate and support the anatomical basis and rationale of the transoral surgery and the need for the removal of the SLSG alone, during the management of PR. The study also takes advantage of the increasing prevalence of plunging ranulas as it was observed in HIV infected-patients to confirm the effectiveness of the proposed surgical technique, including in this particular group of patients.

2. Materials and methods

This retrospective study was based on cases of PR, observed and treated at a tertiary outpatient unit of Maxillo-facial and Oral Surgery. The diagnosis of plunging ranula was based on clinical presentation, magnetic resonance imaging (MRI) investigation and histological confirmation.

Eleven patients diagnosed HIV positive (HIV+), with various clinical degree of PR, underwent surgery using the transoral approach with evacuation of the pseudocyst.

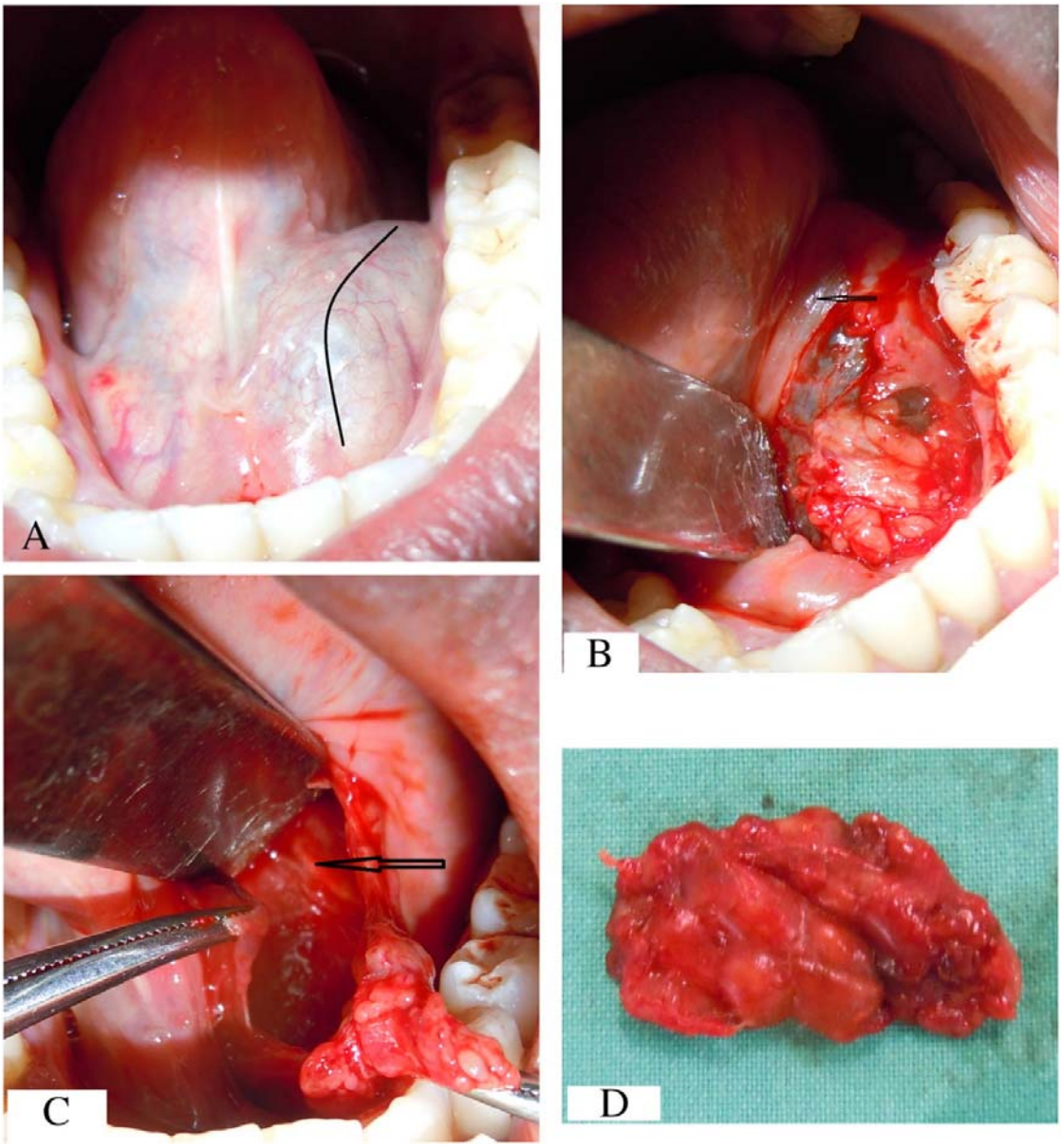

Fig. 1. A. Clinical presentation of the intraoral component of a plunging ranula, with the design of the planned surgical incision (black line)

B. Same patient as in Fig. 1A. After the incision of the oral mucosa layer, the dissection is carried- out laterally towards the body of the mandible. The pseudocyst and the sublingual gland are exposed. The Wharton duct is protected by retracting it lingually (black arrow).

C. Same patient as in Fig. 1A. The excision of the salivary gland and the attached pseudocyst is almost completed. Any dissection of the pseudocystic membrane is absolutely avoided. The lingual nerve can be identified deeper and below the transparent pseudocystic membrane (black arrow).

D. Same patient as in Fig. 1A. The hypertrophic appearance of the sublingual salivary gland after complete excision.

The surgical procedure consisted of the transoral excision of the offending ipsilateral SLSG, with evacuation of the pseudocystic content alone. All the patients were treated as outpatients. The surgical procedures were performed under local anesthesia, irrespective of the size of the extra oral component of the ranula. The inferior alveolar nerve block was performed using lignocaine HCl 2% with adrenaline (1:80000). Special attention was directed towards the effective block of the lingual nerve, either unilaterally or bilaterally, depending on the case. A supplementary injection of the anesthetic solution was injected in the lingual aspect of the mandibular symphysis. The oral mucosa incision was made buccally and parallel to the submandibular salivary gland (SMSG) Wharton duct (Fig. 1A). The pseudocystic membrane was exposed and the SLSG, lying under the submucosal plane was identified (Fig. 1B). The dissection of the SLSG with the attached pseudo cystic membrane was carried-out laterally towards the body of the mandible. Neurovascular structures, especially the lingual nerve, were identified through the partially transparent pseudocystic membrane (Fig. 1C) and protected by retracting them medially towards the tongue. Therefore, there was, no need to dissect and/or identify the lingual nerve and artery [10]. The body of the SLSG with part of the attached pseudo membrane was finally removed (Fig. 1D). The cystic cavity was intraorally evacuated and rinsed with normal saline solution. Any extensive dissection of the pseudocystic membrane, especially towards the lingual side, was absolutely avoided. No drain was placed in the empty pseudocyst at the end of the procedure. The oral mucosa was then loosely sutured with absorbable material (Vicryl® 3/0 or 4/0), paying attention not to damage the Wharton duct. At the end of the surgical procedure, the patency of the Wharton duct was assessed by massaging the ipsilateral SMSG and checking the free flow of the saliva from the opening of the duct. Patients were discharged on the same day after the operation. A prescription of analgesics, nonsteroidal anti-inflammatory drugs and antibiotics was provided. A non-alcohol chlorhexidine-based (0.2%) mouthwash was also prescribed stating from day one postoperatively.

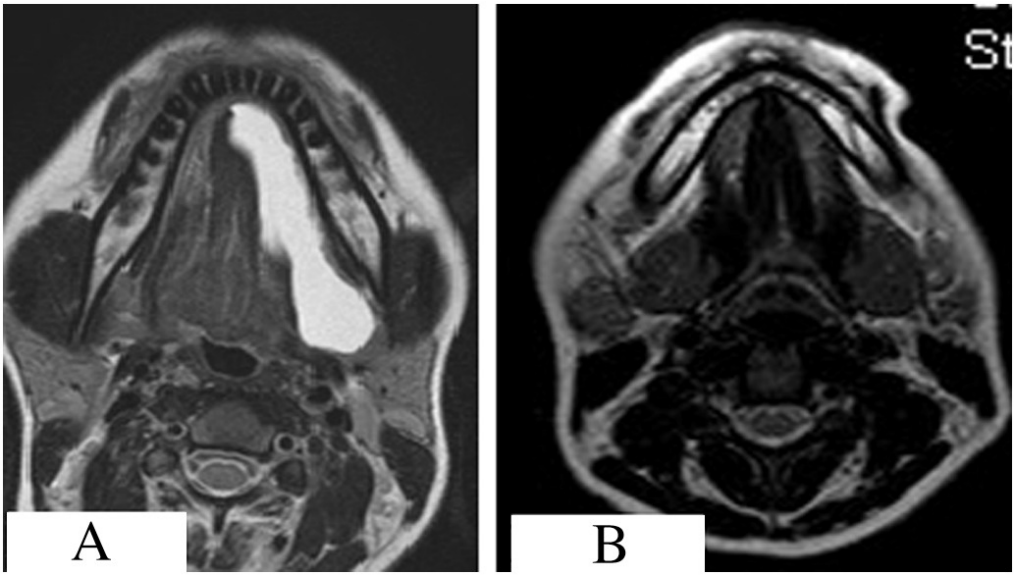

Fig. 2. A. Same patient as in Fig. 1A. Pre-operative MRI investigation (axial view) of the plunging ranula.

B. Same patient as in Fig. 2A. Follow-up MRI investigation (axial view), 4 months postoperatively, showing a complete resolution of the plunging ranula.

The volume and tenderness of the ipsilateral SMSG was also clinically assessed during the first follow-up postoperative visit, five days later. Postoperative clinical examination, including clinical patients' pictures, as well as MRI imaging were used for monitoring purposes. (Fig. 2A, B). The follow-up period ranged from tree to 15 months.

3. Results

Ninty (90) adults presenting with oral mucocele, type ranula were recoded. Seventy (77%) of them were diagnosed with HIV infection. Plunging ranula was recorded in 35 (50%) patients from the latter group (Table 1).

Table 1. Distribution of ranula type in relation to patients' HIV infection status.

Ranula type	**HIV positive**	**HIV negative**	**Total**
Simple ranula[a]	35 (50%)	18 (90%)	53(58.9%)
Plunging ranula[b]	35 (50%)	2 (10%)	37(41.1%)
Total	70 (77.8%)	20 (22.2%)	90

HIV = Human Immunodeficiency Virus.

[a]Oral mucocele in the floor of the mouth, above the mylohyoid muscle.
[b]Oral mucocele in the floor of the mouth extending below the mylohyoid muscle and/or the neck.

Eleven HIV+ patients, presenting with PR, were enrolled in this study and successfully operated using the surgical technique as previously described. The SLSG removed from HIV+ patients appeared macroscopically hypertrophic (Fig. 1D).

In terms of postoperative complications, there was no neurological fallout (e.g., lingual nerve) observed, no obstruction or damage to the Wharton duct observed, and no postoperative hematoma or infection reported. The complete resolution of the plunging part of the ranula was clinically observed in all operated patients.

The postoperative MRI imaging (Fig. 2A, B) confirmed the absence of any form of recurrence of the plunging ranula in the short-term (3 to 5 months) for five patients, and medium-term (15 months) recurrence for the other six patients. The histological results of all samples were reported as sublingual salivary gland associated with a mucus extravasation/retention phenomenon cyst.

4. Discussion

It is commonly accepted that the majority of ranula (simple or plunging) originate from the SLSG [6,23]. While a simple ranula is confined to the supra-mylohyoid muscle space, the PR type may conversely, expend in and/or beyond the infra-mylohyoid muscle spaces, via its posterior boarder or through multiple hiatuses (Fig. 3) described in the body of the muscle to reach in some cases remote areas such as the neck.

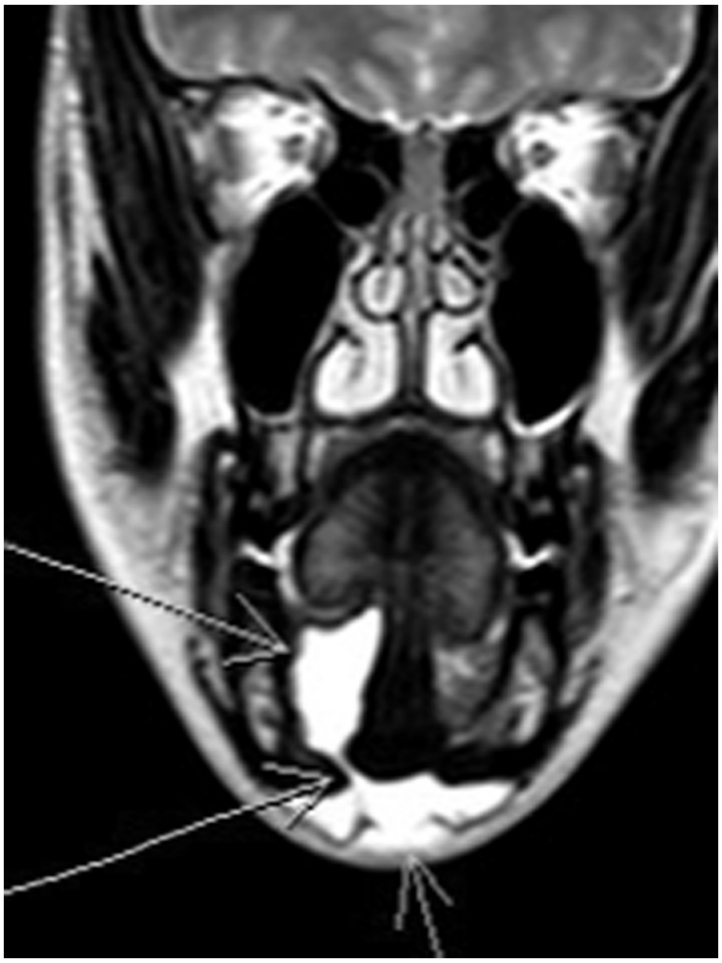

Fig. 3. Coronal view of a plunging ranula with a very tight trans-mylohyoid hiatus. The use of the cervical approach with the subsequent dissection/excision of the ranula may be in this case very laborious.

It is important, from the physio-pathogenesis of PR to note the fact that mucus collection and pseudocyst formation are partly influenced by the gravity factor. This means that the SLSG is often, if not always, found just below the oral mucosa and above the mylohyoid muscle, irrespective of the size and the expansion of the extra oral component (Fig. 1A, B). This anatomical location of the SLSG; just below a single and thin layer of oral mucosa is of great importance and actually, justifies the transoral surgical approach for the excision of the gland. It also makes transoral surgery of the SLSG a relatively simple surgical procedure (Fig. 1A,B, C).

Several and various modalities of treatment for ranula have been described in the literature. These modalities include both non-surgical and surgical methods of treatment. However, the transoral complete excision of the offending ipsilateral SLSG alone, with intraoral evacuation of the pseudocystic content seems to be the most trusted by the majority of surgeons, as the surgical method for the treatment of PR [[5], [6], [7], [8], [9], [10], [11], [12], [13], [14], [15], [16],24]. This study focused on the application of the abovementioned approach for the treatment of the PRs in a specific group of HIV+ patients, presenting with this lesion.

While the transoral surgical approach, has been and is still widely recommended as treatment of choice for PR, there have been, however, some literature reports that continue to described and advocate for debatable surgical practices. The following three questions constitute the core of these controversial surgical practices:

1. What is the rational and need for the dissection and/or excision of the pseudocystic wall of the PR, alone or together with the excision of the SLSG?
2. What is the rational and need for the use of the extra oral surgical approach during the management of PR?
3. What is the need for the placement of a drain (i.e., Penrose) at the end of the operation?

One of the controversial practices mentioned is the necessity of dissecting the pseudocystic wall (ranula) with or without the excision of the offending SLSG [9,11,13,15,16,19,20,22,24].

The physio-pathogenesis and the submucosal anatomical location of the SLSG make the transoral surgery with evacuation of the ranula a more natural approach. It is a common knowledge that the pseudocystic wall of the PR is not an epithelial secreting membrane. Therefore, it does represent no danger for a potential recurrence of the lesion.

From the present authors' experience, the anatomical location of the SLSG, just below a thin layer of oral mucosa (Fig. 1A, B), simplifies the whole surgical procedure. There is neither a need for any studious dissection of the pseudocystic membrane, nor the necessity to explore and identify the neurovascular bundle (Fig. 1C) [10]. These structures were, in this study, easily kept away from the operating field by retracting them medially (lingually).

The second ongoing controversial practice concerns the rationale behind the need for the use of a transcervical approach as reported by some authors [8,9,15,[19], [20], [21], [22], [23]]. This approach is technically difficult due to the number of anatomical structures separating the submental/submandibular skin and the oral mucosa (Fig. 3). Access to the SLSG via the cervical approach requires massive and studious dissection, as well as a painful navigation through the various anatomical layers (skin, fascia, muscles) and careful protection of important anatomical structures (neuro-vascular bundle and salivary gland ducts) [17]. One may easily imagine the amount of dissection work needed to dissect the ranula and access the SLSG via the posterior margin of the mylohyoid muscle, or through a tight and narrow hiatus (Fig. 3). This approach increases the risk and potential for complications both during and postoperatively [7,14,15,22].

The primary source of the formation of the PR remains mainly the SLSG [12]. The size and the location of the pseudocyst play only a symptomatic role in the clinical presentation of the PR [11,15]. The physio-pathogenesis of ranula formation and the sub-mucosal anatomical location of the SLSG are explicit [6,17]. Whatever may be the size of the PR, the real culprit remains the tiny salivary gland, located and easily accessible intraorally. Furthermore, the size of the plunging or the extra oral component of the ranula does not make, necessarily, the intraoral excision of the SLSG a different and major surgical procedure, as illustrated by the present case series.

We therefore, recommend that surgeons resist any temptation to dissect the pseudocystic membrane or to approach the PR via its extra oral component irrespective of what may be its size [1,24].

There are also reports from the literature mentioning the placement of a drain in the empty cystic cavity at the end of the surgical procedure. Some surgeons will communicate the drain to the intraoral cavity [12,16,21,23,24], while others will direct it extra orally through a cutaneous incision [14,23].

The need and attempt to place a drain in the empty pseudocystic cavity seem to arise from the surgeon fear that the cavity may not be completely evacuated and, therefore, a residual amount of fluid may persist. This is far from the reality and the practice only increases the risk of postoperative infection. No drain has been placed at the end of the surgical procedure in this cases series, irrespective of the size and the situation of the extra oral component of the PRs.

Several postoperative complications have been reported in the literature following surgical treatment of ranula, especially the plunging type [7,12,[14], [15], [16]]. The severity of these complications seems to be related to the surgical technique [7].

The most damaging complication, depending on the size of the plunging ranula, is the neurological fallout of the lingual and/or the hypoglossal nerve. Transient facial palsy due to injury to the marginal mandibular branch of the facial nerve has also been mentioned, and is especially associated with the extra oral approach [12].

The obstruction and/or the injury to the salivary gland duct (Wharton duct) is another feared post-operative complication directly related to the surgical technique used [7,14]. The risk of occurrence of injury to the Wharton duct increases with the extent of the dissected field, both from an intraoral and from an extra oral approach. The risk of bleeding during and/or postoperatively may also be higher following an extended dissection.

The increased risk of postoperative complications following a laborious dissection of the ranula, and that following an extra oral approach, makes these surgical practices questionable [[14], [15], [16],19].

The last complication that is often overlooked is the cosmetic dimension. Indeed, the needed large skin incision, followed by massive dissection work during the extra-oral approach, exposes the patient to possible submental / submandibular and cervical scarring [[18], [19], [20],22]. Although this issue may not be as important in male patients, it does, however, require consideration in female counterpart. Nevertheless, this constitutes another positive advantage of the transoral approach. No post-operative complication was observed in this case series for patient with sufficient follow-up time.

The clinical success of the transoral surgical technique as described in this study was confirmed by the absence of any form of cystic content that persisted or developed in the postoperative follow-up period (Fig. 2A, B).

The median time for recurrence of operated (transoral SLSG removal) PR is not well established. Morton et al. [14] reported a median time of 24-month after transoral SLSG excision. However, the author did not provide details about the excision or not of the associated cystic pseudo membrane. Huang et al. [11] reported a recurrence of PR at three months after surgery. The follow-up period in this case series extended from three to 15 months without any recurrence or complication.

The surgical technique described in this study is in agreement with what has already been reported in the literature, about the transoral SLSG removal in HIV-negative patients. The transoral removal of the SLSG with simple evacuation of the ranula content is an effective surgical modality for PR treatment. There is no need for the dissection / excision of the pseudocystic membrane, no need for the use of an extra oral approach, and there is no need for placement of a drain at the end of the surgical procedure.

Nevertheless, one may be concerned about the use of local anesthesia in this study involving PRs in adult patients. From the present authors' experience, the use of local anesthesia is feasible, adequate and safe, especially for those patients with any form of contraindication for general anesthesia (GA). Local anesthesia is also cost effective. Patients underwent surgery as outpatient and were discharged on the same day. It is, however, clear that GA remains, indeed, more comfortable for anxious adults and obviously for pediatric patients.

No postoperative complications were reported in this case series comprising eleven HIV-infected patients. This part of the study simply suggests that HIV-infected patients may be subjected to the surgical treatment of PR similar to any other immunologically competent patients. However, the presentation of multiple oral mucocele lesions in HIV-infected patients and the reported potential beneficial effect of highly active antiretroviral drugs on ranula, may represent limitations to systematic surgical removal of SLSG in this particular group of patients [1,25].

5. Conclusion

Based on the anatomical location of the SLSG in the floor of the mouth, the transoral surgical complete excision of the ipsilateral offending gland, coupled with intraoral evacuation of the pseudocystic content alone, remains anatomically the most rational approach for the management of plunging ranula. There is neither need for ranula dissection, nor rationale for the use of cervical approach. The placement of a drain at the end of the surgical procedure is also unnecessary.

CRediT authorship contribution statement

Kabunda Syebele: Conceptualization, Methodology, Investigation, Writing - original draft.
Thifhelimbilu I. Munzhelele: Methodology, Investigation, Writing - review & editing.

References

[1] Syebele K, Bütow K-W. Oral mucoceles and ranulas may be part of initial manifestations of HIV infection. *AIDS Res Hum Retroviruses* 2010; 26: 1079–1085. https://doi.org/10.1089/aid.2010.0051.

[2] Butt FM, Chindia ML, Kenyanya T, Gathece LW, Rana F. An audit of ranulae occurring with the human immunodeficiency virus infection. *J Oral Maxillofac Pathol* 2010; 14:33–35. (doi:10.4103%2F0973-029X.64312).

[3] Kamulegeya A, Okello SM. Ranulas: possible signs for HIV/AIDS? 1 year Ugandan descriptive study. *Acta Odontol Scand* 2012; 70: 149–153. https://doi.org/10.3109/00016357.2011.600709.

[4] Syebele K, Munzhelele TI. Oral mucocele/ranula: another human immunodeficiency virus-related salivary gland disease? *Laryngoscope* 2015; 125: 1130–1136. https://doi.org/10.1002/lary.25058.

[5] De Visscher JGAM, Van der Wal KGH, De Vogel PL. The plunging ranula pathogenesis, diagnosis and management. *J Cranio-Max-Fac Surg* 1989; 17: 182–185.

[6] Harrison JD. Modern management and pathophysiology of ranula: literature review. *Head Neck* 2010; 32: 1310–20. https://doi.org/10.1002/hed.21326.

[7] Zhao Y-F, Jia J, Jia Y. Complications associated with surgical management of ranula. *J Oral Maxillofac Surg* 2005; 63: 51–54. https://doi.org/10.1016/j.joms.2004.02.018.

[8] Sigismund PE, Bozzato A, Schumann K, Iro H, Zenk J. Management of ranula: 9 years'clinical experience in pediatric and adult patients. *J Oral Maxillofac Surg* 2013; 71: 538–544. https://doi.org/10.1016/j.joms.2012.07.042.

[9] Chena JX, Zengaa J, Emericka K, Deschlera D. Sublingual gland excision for the surgical management of plunging ranula. *Am J Otolaryngol* 2018: 497–500.

[10] Wood G, Syyed N, Wales C. Sublingual gland excision: a dissection carried out following adjacent anatomical structures. *Br J Oral Maxillofac Surg* 2016; 54: 927–929. https://doi.org/10.1016/j.bjoms.2016.06.014.

[11] Harrison JD, Kin A, Al-Ali S, Morton RP. Postmortem investigation of mylohyoid hiatus and hernia: aetiological factors of plunging ranula. *Clin Anat* 2013; 26: 693–699.

[12] Huang SF, Liao CT, Chin SC, Chen IH. Transoral approach for plunging ranula –10-year experience. *Laryngoscope* 2010; 120: 53–57. https://doi.org/10.1002/lary.20674.

[13] Zhao YF, Jia Y, Chen XM, Zhang WF. Clinical review of 580 ranulas. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2004; 98: 281–287. https://doi.org/10.1016/j.tripleo.2004.01.013.

[14] Samant S, Morton RP, Ahmad Z. Surgery for plunging ranula: the lesson not yet learned? *Eur Arch Othorinolaryngol* 2011; 268: 1513–1518. https://doi.org/10.1007/s00405-011-1509-y.

[15] Patel MR, Deal AM, Shockley WW. The oral and plunging ranulas: what is the most effective treatment? *Laryngoscope* 2009; 119: 1501–1509. https://doi.org/10.1002/lary.20291.

[16] Yang Y, Hong K. Surgical results of the intraoral approach for plunging ranula. *Acta Otolaryngol* 2014; 134: 201–205. https://doi.org/10.3109/00016489.2013.831481.

[17] Netter FH. *Atlas of human anatomy*. 6th ed Philadelphia: Saunders; 2014. [plates 43, 46].

[18] Kolomvos N, Kalfarentzos E, Papadogeorgakis N. Surgical treatment of plunging ranula: report of three cases and review of literature. *Oral Maxillofac Surg Cases* 2019; 5: 100098.

https://doi.org/10.1016/j.omsc.2019.100098.
[19] Daniel Kokong D, Iduh A, Chukwu I, Mugu J, Samuel Nuhu S, Augustine S. Ranula: current concept of pathophysiologic basis and surgical management options. *World J Surg* 2017; 41: 1476–181. https://doi.org/10.1007/s00268-017-3901-2.
[20] Olojede ACO, Ogundana OM, Emeka CI, Adewole RA, Emmanuel MM, Gbotolorun OM, et al. Plunging ranula: surgical management of case series and the literature review. *Clinical Case Reports* 2018; 6: 109–114. https://doi.org/10.1002/ccr3.1272.
[21] Abdul-Aziz D, Adil E. Ranula excision. *Oper Tech Otolaryngol Head Neck Surg* 2015; 26: 21–27. https://doi.org/10.1016/j.otot.2015.01.005.
[22] Nilesh K, Malik NA, Patil P, Chapi MD. Large plunging ranula presenting as isolated neck swelling: steps in diagnosis and surgical steps in management. *J Clin Diagn Res* 2015; 9: MD01–3. https://doi.org/10.7860/jcdr/2015/12419.6016.
[23] Lesperance MM. When do ranulas require a cervical approach? Laryngoscope 2013; 123: 1826–1827. https://doi.org/10.1002/lary.23937.
[24] Karino M, Kanno T, Iwahashi T, Ide T, Kaneko I, Yoshino A, et al. A rare case of plunging ranula with local recurrence and wide spread to the infratemporal fossa treated successfully by an intraoral surgical treatment. *J Oral Maxifac Surg Med Pathol* 2017; 29: 240–244. https://doi.org/10.1016/j.ajoms.2016.12.001.
[25] Syebele K, Munzhelele TI. The potential impact of highly active antiretroviral therapy on the treatment and epidemiology of ranula in human immunodeficiency virus-positive patients. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2013; 116: e32–36. https://doi.org/10.1016/j.oooo.2013.02.016.