

Case Report

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Plunging Ranula as Initial Manifestation of HIV-AIDS: A Case Report and Literature Review

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Abstract

**Introduction:** Ranulas are defined as the extravasation of mucus into an intraoral cystic cavity produced by an injury to the excretory ducts or the acini of the sublingual gland. Plunging ranulas are generated when the salivary collection penetrates the mylohyoid through a dehiscence of its fibers, invading the submandibular space. The close relationship between these lesions and HIV-AIDS infection has been reported since 2004, with a 75.88% rate of positive cases. The objective of this article is to present the management of patient with a plunging ranula, which made it possible to address the diagnosis of HIV-AIDS.

**Case Report:** A 28-year-old male patient presented with a painless and fluctuating swelling in the laterocervical space and the floor of the mouth, which produced dysphagia and dyslalia. Serological tests for HIV were carried out and had a reactive result. Surgical treatment was performed via intraoral approach, with a favorable evolution at 12 months of follow-up.

**Conclusion:** Ranulas, and particularly plunging ranulas, should be considered in the group of oral lesions associated with HIV-AIDS infection, as they may even be the initial manifestation of the disease.

**Keywords:** HIV, Oral manifestation, Plunging ranula, Transoral surgery

Introduction

In 2017 the World Health Organization described the term “ranula” as a mucous extravasation into an intraoral cystic cavity which is generally associated with the rupture of the excretory ducts or the acini of the sublingual gland (SLG) [1]. A ranula (from Latin rana) is so named because it clinically resembles the swollen abdomen of a frog [2,3].

According to the site of the primary swelling, ranulas are classified into two clinical types: (a) superficial or oral lesions are masses confined to the floor of the mouth above the mylohyoid muscle, which are painless, fluctuating, balloon-like, bluish, variable in size; they may superiorly and medially displace the tongue, involving disorders in speech and swallowing; and (b) deep lesions, also known as cervical or plunging ranulas (PR), are painless cervical masses, variable in size, which occupy the submandibular triangle and may coexist with an intraoral ranula; PRs amount to

7-30% of the whole universe [1-3]. A cervical ranula originates in the herniation of the mucous content and, occasionally, in the sublingual gland on the mouth floor through a mylohyoid muscle dehiscence, or the posterior rim of the muscle into the submandibular or submental space [1,2,4-7].

There is a slight epidemiological predominance of oral ranulas in women, whereas PRs are more frequent in male patients [2,4]. PRs are prevalent in young individuals in the first three decades of life [2,4,8]. The literature shows that its total incidence rate is 2.4-2.6 over 100,000 inhabitants [9]. Frequency of occurrence is equivalent on both sides of the mouth although ranulas may appear bilaterally [5,9].

As in the case of labial mucoceles, histology shows that a PR is a collection of saliva, surrounded by inflammatory fibrous granulation tissue forming a histiocyte-rich capsule, which appears as a

pseudocyst without epithelial lining (Figure 3c, d) [1,3,6].

Although diagnosis is mainly performed through clinical examination, diagnostic ultrasound imaging, computed tomography (CT) and magnetic resonance imaging (MRI) prove useful to determine the size and location of the ranula and its relationship with the compromised SLG [2,10].

The treatment options advocated by different authors for the management of this injury are aspiration, sclerotherapy, marsupialization, or the excision of the ranula exclusively. The recurrence rate has been 57–73% in the population of patients treated with these techniques [2,6]. The gold standard procedure is the surgical removal of the SLG and draining its collection with or without the excision of its capsule, with a recurrence rate being 0–1.5% [2,4-6,11,12]. The literature shows that the PR has traditionally been approached cervically; however, in the last decade publications show a trend in favor of the transoral approach as the treatment aims at eliminating the compromised SLG, the cause of the disorder, which is done intraorally for ease of accessibility [5,6,11,12].

The main etiological factor in the onset of this injury is trauma, which may occur on the floor of the mouth, usually associated with dental treatments, or in the cervical area [1-3]. In 2004 Chizonga and Rusakaniko referred to the rising prevalence of oral ranulas together with and HIV/AIDS in Zimbabwe [13]. The study provided evidence that the percentage of HIV/AIDS patients with oral ranulas was significantly higher than the percentage of HIV/AIDS patients in the general Zimbabwean population. On the other hand, the authors show a higher progressive incidence of ranulas as of 1999, especially in pediatric patients, as well as Kaposi's sarcoma (KS) in adults [4,13].

Numerous articles by African authors, mainly Syebele and Munzhelele, have ensued showing the relationship between ranulas and HIV/AIDS, some of them arguing that this injury could be an initial manifestation of the disease [5,8,14].

The aim of this article is to present the management of a patient with PR, which resulted in the diagnosis of HIV/AIDS.

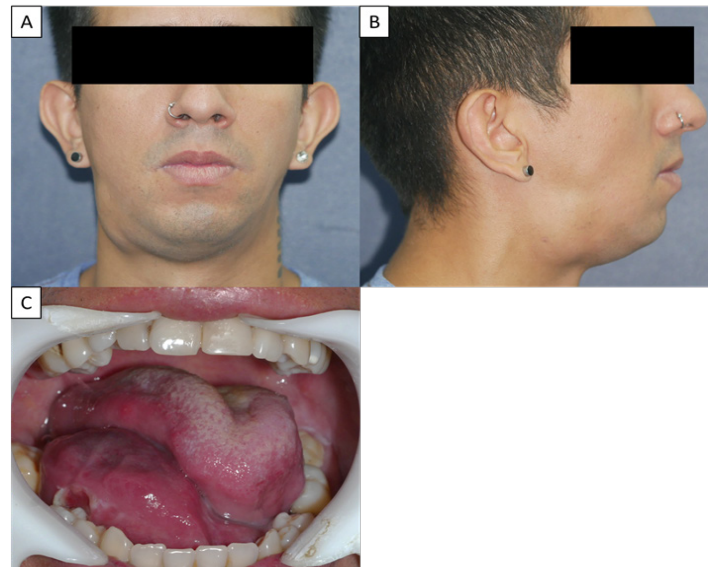
### Case Report

A 28-year-old Caucasian male presented to the Department of Oral and Maxillofacial Surgery, Gral Manuel Belgrano Acute Hospital, San Martín District, Buenos Aires, Argentina, with a 2-month history of painless swelling in the right latero-cervical region and homolateral floor of the mouth (Figure 1).

During anamnesis, the only positive finding was a 5 pack-year history of smoking. The patient had experienced a spontaneous increase of volume on the floor of the mouth, on the right region, the size of which was not constant. He eventually developed a latero-cervical collection with similar characteristics.

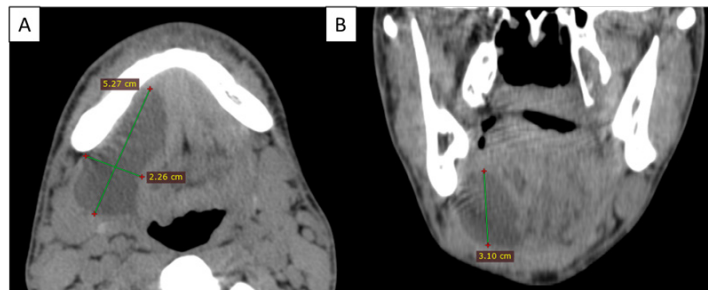
On examination, a painless increase of cervical volume with fluctuant consistency and normal-looking overlying skin was apparent on the right region, levels IA and B, (Figure 1 A-B). Intraorally, the patient had a homolateral lesion on the floor of the mouth, which was balloon-like, with fluctuant consistency, painless, bluish, with a lateral ulceration on the surface (Figure 1 C). His tongue showed

cephalic and medial displacement causing dysphagia and dyslalia.



**Figure 1:** A: Frontal view. B: Right lateral view. C: Intraoral view

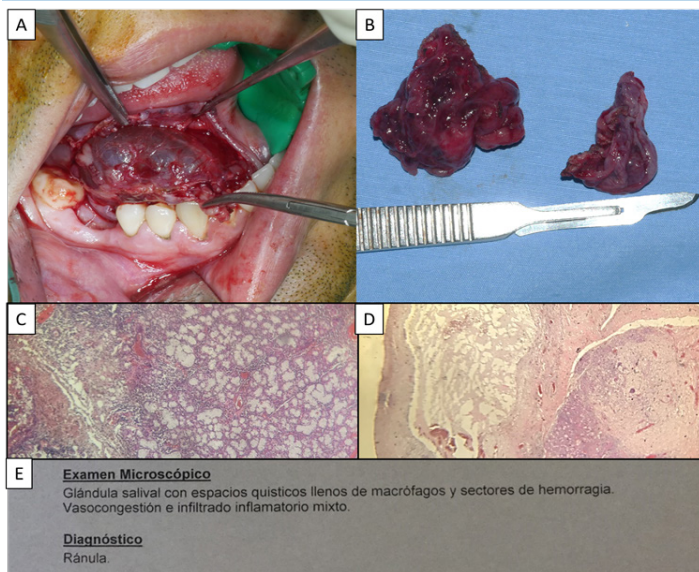
Head and neck CT scans demonstrated the presence of a hypodense image in the right sublingual and submandibular space, lateral to the geniohyoid and genioglossus muscles, 5.27x3.1x2.26 cm in size. The submandibular gland was compressed by the collection while the mylohyoid muscle was crossed by the ranula (Figure 2).



**Figure 2:** CT of head and neck. A: Axial view. B: Coronal view

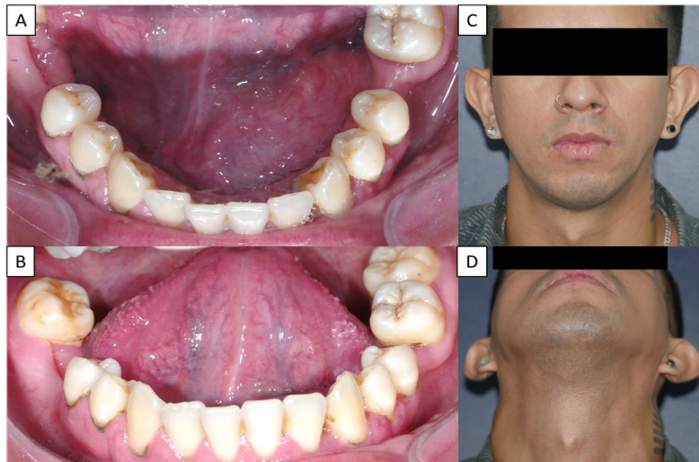
Consent was given by the patient for the performance of a chemiluminescence assay to detect HIV Ag/Ac plasma; reactive results were confirmed by a second test. The patient was referred to the hospital Infectious Diseases Department, which reported a CD 4 lymphocyte count and a viral load of 94 cells/mm<sup>3</sup> and 84 copys/mL respectively. Highly active antiretroviral treatment was initiated with efavirenz 600mg, emtricitabine 200mg and tenofovir disoproxil 245 mg (Atripla)

Surgery was administered under general anesthesia, using the intraoral approach to the floor of the mouth from the ostium towards the mesial aspect of mandibular first molar (Figure 3 A). The capsule was dissected identifying medially the lingual nerve and the Wharton duct. Complete excision of the right SLG, drainage of saliva collection and partial removal of the capsule were performed (Figure 3 B). The specimen was submitted for surgical pathology analysis, which confirmed the diagnosis of ranula (Figure 3 C, D, E).



**Figure 3:** A: Transoral surgical approach. B: Macroscopic sublingual gland and capsule. C-D: H-E stained histopathologic images. E: Pathology analysis “Salivary gland showing cystic spaces full of macrophages and hemorrhages sectors. Vasocongestion and mixed inflammatory infiltrate”

Immediate postoperative controls and regular follow-up visits showed a favorable evolution with neither recurrence nor motor or sensitive tongue compromise after 12 months had elapsed (Figure 4).



**Figure 4:** A-B: Postoperative intraoral images after 12 months. C: Postoperative frontal image after 12 months. D: Postoperative cephalocaudal image after 12 months

## Discussion

A PR is a rare injury with a prevalence of 0.175-0.75 cases over 100,000 inhabitants. Despite its low frequency of occurrence, its etiopathogenesis has been extensively studied [5-7]. The theory most widely postulated is that PR is secondary to a simple ranula, caused by the rupture of the Rivinus ducts or the acini of the SLG. This lesion in turn results in the extravasation of saliva into the floor of the mouth, and then through the mylohyoid muscle due to the dehiscence of its fibers, or through its posterior rim with the usual invasion of the submandibular triangle. Thus, the patient clinically reports a painless and fluctuant increase of volume, located on the floor of the mouth and the homolateral submandibular space. There are also reports of synchronous bilateral plunging ranulas [5,9]. Although this injury is painless, it may cause speech and swallowing disorders and even a partial upper airway obstruction. Hence, treatment of these patients should not be delayed.

This saliva collection is surrounded and encapsulated by inflammatory fibrous granulation histiocyte-rich tissue. Treatment should focus on removing the affected SLG, as the cause of the injury, and draining the fluid. It is not mandatory to perform the excision of the fibrous capsule as it is not apt to produce recurrence. This is the reason why the latest publications quote a preference for the surgical treatment by intraoral approach, the recurrence rate being 0–1.5% [5,6,11,12]. The SLG is anatomically located on a submucosal plane on the floor of the mouth, lined only by a thin layer of oral mucosa. The cervical approach involves invasion of the submandibular space through the superficial cervical aponeurosis, and excision of the SLG through the submandibular hiatus, this being a procedure with greater morbidity, increasing its recurrence [5,11,12].

SLG trauma, either intraoral or cervical, has been classically considered the pathognomonic etiologic factor of simple and plunging ranulas. Intraoral trauma is usually associated with dental iatrogenic procedures.

Since 2004 there have been several articles in the literature, particularly by African authors, expounding the close relationship between ranulas and HIV. The first authors to describe this finding are Chizonga MM from Zimbabwe, followed by Butt FM from Kenya, Syebele K from Sud Africa, and Kamuelegeya A from Uganda, who reported cases of patients with ranulas and HIV (Table 1) [4,5,8,13,15,16].



**Table 1: HIV+ patients with ranulas in scientific publications**

Author	Year	Country	Ranulas (total)	% HIV+ (n)
Chidzonga MM	2004	Zimbabwe	26	88.50 (23)
Butt F	2010	Kenya	28	67.90 (19)
Syebele K	2010	South Africa	50	68.75 (34)
Kamulegeya A	2011	Uganda	57	73.70 (42)
Syebele K	2015	South Africa	89*	78.65 (70)
Syebele K	2019	South Africa	90	77.80 (70)
TOTAL			340	75.88 (258)

\* Only ranulas are included. Labial and lingual mucoceles are excluded.

It should be noted that in our case, as in numerous others, the ranula was the first manifestation of HIV infection in patients who ignored their health status. Another point to be considered in these publications is that the percentage of HIV increases considerably in PR patients. Syebele K, et al. inform in an article in 2019 that over 37 patients with PR, 35 were HIV+ (94.59 %) [5].

According to UNAIDS data, the African continent bears the highest incidence and prevalence of HIV AIDS cases in the world. Consequently, the manifestations of the disease published by African practitioners have considerable weight as their observations are to be replicated on the other continents. Knowledge of the oral manifestations of HIV AIDS is a matter of paramount importance

in the dental and medical practice because these findings may accelerate the diagnosis of the disease and its treatment, eventually reducing the dissemination of the infection.

In 1991, practically a decade after HIV was discovered as the causative agent of AIDS, an update of the classification, and diagnostic criteria of oral lesions in HIV infection was published [17]. Three groups of orofacial lesions were identified: in Group 1 there were lesions strongly associated with HIV infection; in Group 2, lesions less commonly associated with HIV infection; and in Group 3, lesions rarely seen in HIV infection (Table 2). There were further updates and reviews of the oral manifestations associated with HIV AIDS, but ranulas are not considered in none group yet [18-22].

**Table 2: Oral lesions associated with HIV/AIDS**

Lesions strongly associated with HIV <sup>o+</sup>	Lesions less commonly associated with HIV <sup>o+</sup>	Lesions rarely seen in HIV <sup>o+</sup>
Candidiasis (Erythematous and Pseudomembranous)	Bacterial infections (Mycobacterium avium intracellulare and Mycobacterium tuberculosis)	Bacterial infections – Actinomyces Israel – Escherichia coli – Klebsiella pneumoniae
Hairy leukoplakia•	Melanotic hyperpigmentation	Cat scratch disease
Kaposi's sarcoma	Necrotizing ulcerative stomatitis	Erythema multiforme
Non Hodgkin's lymphoma	Salivary gland disease (xerostomy, inflammation of the major salivary glands)	Epithelioid (bacillary) angiomatosis
Periodontal disease (Linear gingival erythema, Necrotizing (ulcerative) gingivitis, Necrotizing (ulcerative) periodontitis)	Thrombocytopenic purpura	Neurologic disturbances – Facial palsy – Trigeminal neuralgia
	Ulceration NOS (not otherwise specified)	Fungal infection other than candidiasis – Cryptococcus neoformans – Geotrichum candidum – Histoplasma capsulatum – Mucoraceae (mucormycosis/zygomycosis) – Aspergillus flavus
	Viral infections (Herpes simplex virus, Human papillomavirus, Condyloma acuminatum, Focal epithelial hyperplasia, Verruca vulgaris, Varicella zoster virus, Herpes zoster, Varicella)	Recurrent aphthous stomatitis

		Drug reactions (ulcerative, erythema multiforme, lichenoid, toxic epidermolysis)
		Viral infections – Cytomegalovirus – Molluscum contagiosum

As proponents of Dr. Syebele's, et al. viewpoint, our team believe that ranulas, and PRs in particular, should be considered as a lesion associated with HIV infection, among those of the salivary gland diseases in Group 2, and even as the first manifestation of the disease. This article intends to add evidence for future reviews.

### Conclusion

As advocated in the series of cases published in Zimbabwe, Kenya, South Africa, and Uganda, we believe that ranulas, and plunging ranulas in particular, may be oral lesions associated with HIV/AIDS, or even the initial manifestation of the disease. Our position is that serological tests should be performed to detect HIV in all patients with these lesions, so as to publish multicentric results to enrich scientific evidence.

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