

A POST-TRAUMATIC GIANT SUBLINGUAL RANULA

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ABSTRACT

The ranula is a retention cyst that is very thin-walled and odd-looking, which usually appears on the anterior part of the mouth floor. These rare sialoceles appear as bluish, transparent, smooth-surfaced, bustling and painless masses. In this article a 13 year-old girl with sublingual ranula, which occurred after a sublingual injection in the course of the dental procedure, was presented. The patient was referred to us due to her difficulty in swallowing and speaking, and a terrible image of her oral lesion. The diagnosis of

the sublingual oral ranula was made with the typical history and findings of clinical examination and the lesion was treated with total surgical excision. The histopathological examination showed a pseudocyst which had no lining epithelium and had mucinous content. The patient was presented due to the rarity of this size of the simple ranula, and the fact that it has not been reported in neither dermatology nor family medicine literature previously.

Keywords: Ranula, cyst, sublingual, oral mucosa
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POST-TRAVMATİK DEV SUBLİNGUAL RANULA

ÖZET

Ranula ağız tabanı ön bölümünde görülen çok ince duvarlı, nadir ve tuhaf görümlü retansiyon kistidir. Bu çok nadir siyaloseller, mavimsi renkli, şeffaf, düz yüzeyli, hareketli ve ağrısız kitleler olarak belirtilir. Burada polikliniğimize, diş tedavisi sırasında sublingual bölgeye yapılan bir enjeksiyondan sonra gelişmiş, sublingual ranulalı 13 yaşında bir kız çocuğu sunulmuştur. Hasta bize, ağızındaki lezyonun korku-

tucu görünümü, konuşma ve yutma güçlüğü şeklindeki yakınmalarla başvurmuştur. Hastanın tipik hikayesi ve klinik muayene bulguları ile sublingual ranula tanısı konan lezyon total cerrahi eksizyon ile çıkarılmıştır. Lezyonun histopatolojik incelemesinde gerçek döşeyici epitel içermeyen ve müsinöz sıvı ihtiva eden psödokist yapısı izlendi. Olgumuz bu büyüklükteki basit ranulaya nadir rastlanması ve hem dermatoloji ve hem de aile hekimliği literatürlerinde daha önce yer almaması nedeniyle sunulmaktadır.

Anahtar Kelimeler: Ranula, cyst, sublingual, oral mucosa
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INTRODUCTION

Benign cysts of salivary glands which contain mucin are called mucocoeles. In general, mucocoeles are classified into 3 groups.

- 1- Superficial mucocoeles located immediately beneath the mucosa.
- 2- Classical mucocoeles which localize in the upper submucosa.
- 3- Deep mucocoeles which localize in the lower cornium.

According to the rates of incidence, mucocoeles occur most often in the lower lip, followed by lingual, floor of mouth and buccal mucosa. Ranulas are rare, deep mucocoeles that occur in the floor of the mouth.¹ These cysts occur due to either mucus extravasation resulting from rupture of the salivary gland or mucus retention resulting from obstruction of this duct.^{2,3}

CASE

A 13-year-old girl was admitted to the dermatology clinic of our hospital with an oral lesion. She complained difficulty in swallowing and speech, and a frightening appearance as well. In her medical history, she had received a local anesthetic injection in the sublingual area to anesthetize the treatment area by a physician whom she visited for dental treatment 10 days ago. The patient postulated that she felt a burning pain during the injection, and that 4 hours after the treatment, there was initially a dark reddish discoloration and swelling in the injection area, which rapidly grew within 8 hours. The dermatological examination of the oral mucosa revealed a dark blue, semi-translucent, shiny-surfaced, cystic lesion. The lesion was soft, non-pitting and fluctuant to palpation. The shape and size of the lesion changed with the movements of the tongue. When the tongue was moved to the right, the lesion filled the left side of the oral cavity, and reached its largest size which was 7.5x4.5 cm (Figure 1a, 1b).

The bottom of the lesion was soft and there was no induration in the floor of the mouth. The patient had difficulty in speaking and swallowing due to the size of the lesion. There were no additional symptoms. The other areas of the oral mucosa were intact. No regional lymphadenomegaly was detected. No pathologies in the other dermatological and systemic examinations were found. Routine hematological and biochemical tests including INR, PT, APTT and the number of platelets were within normal limits. Based on the patient's typical history and clinical examination, the lesion was diagnosed as a posttraumatic simple sublingual ranula. Thereafter, the cyst was removed by total surgical cystectomy under local anesthesia and sedation with transoral approach. The content of the lesion was hemorrhagic, sticky and viscous. The histopathological examination revealed that the wall of the cyst was fibrotic, and had fresh bleeding areas and many artery sections. There was no true lining epithelium on the surface of the cyst (Figure 2a, 2b). The patient did not have any post-surgical complications. On the 2 weeks (Figure 1c) and 3 months follow up checks, no recurrence of the lesion was found.

DISCUSSION

Ranulas are rare sialocels of the oral mucosa.⁴ They often are localized in the lateral portion of the buccal mucosa.⁵ They usually look transparent, round-oval, planiform and bluish in color.^{4,6} Superficial ranulas appear rather translucent or bluish, while deeper ones may appear in the normal color of the mucosa, or red due to bleeding into the cyst.¹ Due to their appearance, lesions are called "ranula=froglet", which was derived from the Latin word "rana=frog", because of their similarity to the abdomen of a frog.^{3,5,7} Their prevalence is 0.2 in 1000.^{5,7,8} Lesions, are usually unilateral, and 2-3 cm in size.^{9,10} Their incidence is slightly higher in women than in men.^{11,12} Certain types of ranulas have been reported more often in some ethnic groups, →

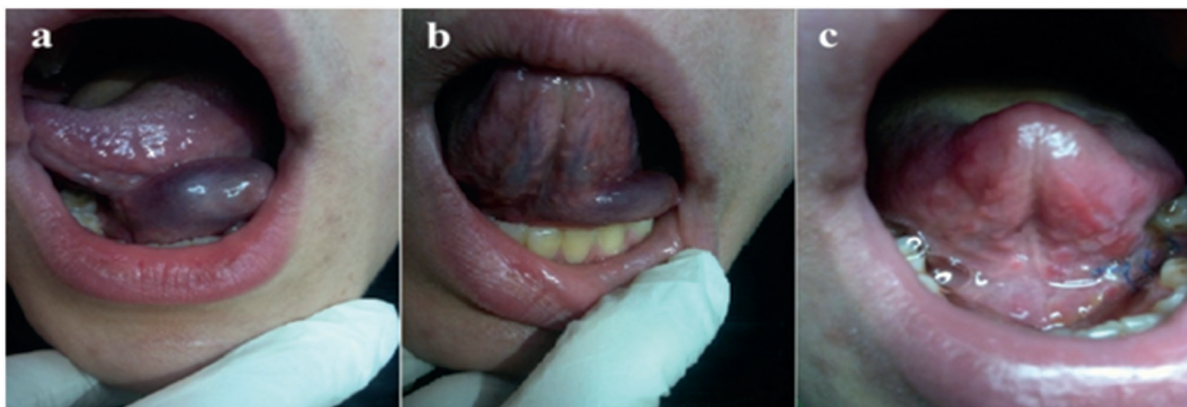


Figure 1(a): Lesion becoming larger and more evident with rightward movement of the tongue.

Figure 1(b): Waned appearance of the lesion at full dorsiflexion of tongue.

Figure 1(c): Post-operative appearance of the lesional zone.

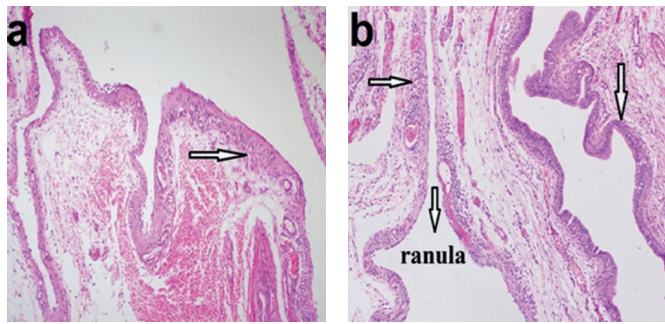


Figure 2(a): Fibrotic cystic wall containing fresh bleeding sites and numerous artery sections. There is not any lining epithelium on the surface of the cyst (HE x100).

Figure 2(b): Ranula wall not containing true lining epithelium (arrow on the left). Respiratory epithelium adjacent to cystic wall (arrow on the right) (HEx100).

such as Pacific Polynesians and Maoris.^{5,13} Although they can occur at any age starting from birth, the lesions occur mostly in the second decade.^{4,6} In the cases reported so far, the age range has been reported as 2 to 61.¹² 75% of the patients were under the age of 20.¹ Ichimura et al. presented their cases as having an age-range of 3-42, and the mean age as 21.5 years.¹⁴ Mizuna and Yamaguchi had cases with an age-range of 4-37, with a mean age of 17.5.¹⁵ Our case was a 13 year-old which complied with the reported age-range in literature. Morphological characteristics of the lesion were similar to those reported in literature. The lesion in our patient was significantly larger than those reported in the literature. The etiologies of the ranulas were not exactly known. However, it has been reported that congenital abnormalities (ductus agenesis or hypoplasia, etc.), trauma or inflammation of the sublingual gland or obstruction of the gland duct by stones of the salivary gland (sialithiasis) may be responsible for the development of the clinical presentation.^{6,12,16-18} Ranulas usually occur as a disease of the sublingual gland. Less frequently, they may stem from the submandibular gland, or rarely from the minor salivary gland.^{2,8,17} Pathogenetically, the ranulas can form as a result of either a mucus extravasation following a sublingual gland duct rupture, or a mucus retention following an obstruction of the duct.^{1-3,5,7} Extravasation cysts seen observed more often than retention cysts.¹⁷ It has been reported that the reaction created by mucus flowing into the soft tissue around the gland due to injury, can be responsible for the pathogenesis as well.^{6,12} The lesion in our case was developed following an injection in the sublingual area. Our patient indicated that she felt a burning pain during the procedure and the lesion developed acutely within 4 hours. Therefore, we thought that similar to the traumatic cases in literature, the lesion of our patient resulted from the traumatization of the gland duct, which led to the salivary gland content leaking under the tissue.

Anatomically, ranulas are classified into 3 groups: Type 1- Simple ranulas developing on the mylohyoid muscle (sublingual). Type 2-Ranulas developing from the back part of the mylohyoid muscles to the cervical area (cervical-plunging ranulas). Type 3-Mixed type (sublingual-plunging).^{2,7,12,15} The lesion in our case localized in the upper part of the mylohyoid muscle, and therefore was considered a simple sublingual ranula.

Although the literature included cervical ranulas that created large masses which led to severe symptoms of pressure, a simple ranula as large as the one found in our case has rarely been reported. Patients with ranulas which are usually painless masses, usually have complains of pressure, difficulty of swallowing or speeching and fullness of the mouth.^{2,5}

Lesions in neonates may lead to difficulty in sucking and feeding.⁴ Large ranulas may even lead to difficulty in breathing.¹⁹ Our patient complained about the feeling of fullness and pressure in the mouth, difficulty in swallowing and speeching, as well as frightening appearance of the lesion.

The diagnosis of a ranula is essentially made on the basis of clinical aspects.² In lesions with a suspected diagnosis, computed tomography and magnetic resonance or other imaging methods can be used, or the biochemical content of the cyst can be analyzed (it contains high levels of amylase and protein).¹²

Ranulas may be histopathologically classified into 2 groups. Retention cysts, which result from ductal obstruction, have a true cystic wall covered with epithelium while extravasation cysts, which result from ductal injury and extravasation are pseudocysts because they do not have a true cystic wall covered with epithelium.^{1-3,20} Most ranulas are pseudocystic.¹⁷ Pseudocystic ranulas consist of a false cystic wall comprised of a loose and vascularized connective tissue and a cavity containing mucin.^{6,13} The histopathological examination of our case revealed that the fibrotic cystic wall consisting fresh bleeding areas and numerous artery sections. The absence of a true epithelium surrounding the cyst, supported the idea that it was a pseudocyst resulting from trauma.

Lingual varices, hemangioma, lymphangioma, branchial cysts, dermoid-epidermoid inclusion cysts, thyroglossal cysts, Wharton duct cysts, palate infections and callosities, lipoma and pleomorphic adenoma, submandibular sialadenitis should be considered in differential diagnoses.^{1,5,17} These diagnoses were ruled out since the lesion in our →

case developed acutely which occurred shortly after an intraoral intervention and had a typical clinical appearance and a typical histopathology.

The most effective treatment of ranula is surgery, but laser ablation, cryotherapy, cauterization, intracystic injection with botox or streptococcal preparation OK-432 are other available options.^{2,5,7} The recurrence ratios following different surgical methods are as follows: 71-100% for incision and drainage, 0-25% for single excision, 61-89% for single marsupialization, 0-12% for marsupialization with packing, and 0-2% for excision of sublingual gland with ranula. A follow-up period may be considered for pediatric cases, due to the possibility of spontaneous resolution of the lesions.²

CONCLUSION

Previous ranula reports were published in oropharyngeal, maxillofacial and dental surgery literature. To the best of our knowledge, there has not been a ranula case reported in dermatology and family medicine literature up to now. On the other hand, simple ranulas of this size in the oral mucosa are very rare. Our case is presented to draw the attention of dermatology and family medicine specialists to these unusual lesions, which have a completely benign nature and a simple and favorable treatment despite their exaggerated clinical appearance.

* The authors declare that there are no conflicts of interest.



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• *Yazılı hasta onamı: Olgumuzdan hastalığı ile ilgili bilgi ve görüntülerin bu bildiriye kullanılabilmesine ilişkin yazılı onam alınmıştır.*