

Huge Plunging Ranula

B. Venkat Suresh · Sambhav K. Vora

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Introduction

Ranula is a form of mucocele which specifically occurs in the floor of the mouth in association with the ducts of the submaxillary or sublingual gland. The term Ranula is derived from the Latin word *Rana* which means ‘Belly of frog’, because the lesions in the floor of the mouth resemble the bulging underbelly of a frog. The formation of ranula is thought to be due to the excretory duct rupture followed by extravasation and accumulation of saliva into the surrounding tissue. The accumulation of mucous in the surrounding connective tissue forms a pseudocyst that lacks an epithelial lining. Ranulas have classically been divided into simple and diving/plunging/cervical. Simple ranulas remain confined to the sublingual space, whereas diving ranulas extend beyond it [1]. Plunging ranula though rare are well documented causes of neck swelling. It is accepted that they arise as a result of extravasation of saliva from the sublingual gland through a hiatus in the mylohyoid muscle. If the only presentation is a cervical swelling, diagnosis can be difficult and advanced diagnostic technique like CT plays an important role in diagnosis.

Case Report

A 23 year old female patient referred by the physician, came with the complaint of mild difficulty while

swallowing. On examination patient was asymptomatic, except for short and obese neck, no abnormality of the floor of the mouth, jaws, teeth or major salivary glands could be detected. Routine blood investigations and thyroid profile was normal. A CT scan of the neck was made, which showed an oval shaped cystic lesion measuring $4.9 \times 3.7 \times 5.1$ cm in midline, in the floor of the mouth extending bilaterally from chin to the hyoid bone region superoinferiorly in the anterior supra hyoid region and mediolaterally extending in between two submandibular triangles (Fig. 1). The mass shows thick capsule not showing much enhancement with I.V contrast and no calcifications were seen (Fig. 2). Rest of soft tissue planes and mandible appeared normal with no evidence of destruction. It was properly differentiated from the other swellings of the anterior neck region and provisional diagnosis of plunging ranula was made. Excision of the mass was carried out under general anesthesia via an intra oral technique approaching floor of the mouth. Careful precautions were taken not to injure vital structures located in the floor of the mouth, the whole mass was excised and sent for histopathological examination. Histopathological examination of the excised mass confirmed the diagnosis of mucous extravasation cyst. Post operative review was done 1 year after the procedure and CT scans were made that time, which revealed normal anatomical resuscitation of the involved area (Figs. 3a, b; Table 1).

Discussion

The term ranula describes a blue translucent swelling in the floor of the mouth reminiscent of the underbelly of a frog. Hippocrates described ranulas as secondary to inflammation. The occurrence of the ranula is rare and the reported

B. V. Suresh (✉) · S. K. Vora
Department of oral and maxillofacial surgery, Gitam dental college and hospital, Visakhapatnam 530045, Andhra Pradesh, India
e-mail: drvenkatsuresh@gmail.com

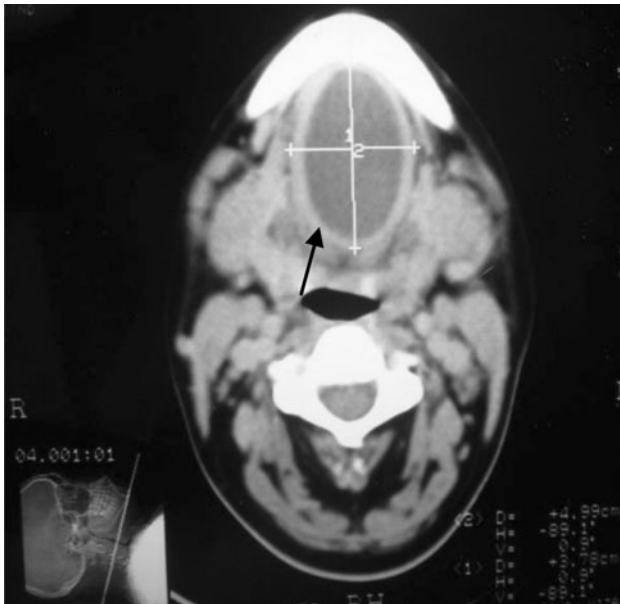


Fig. 1 Pre operative CT scan showing oval shaped cystic mass lesion measured $4.9 \times 3.7 \times 5.1$ cm in midline, in the floor of the mouth extending bilaterally from chin to the hyoid bone

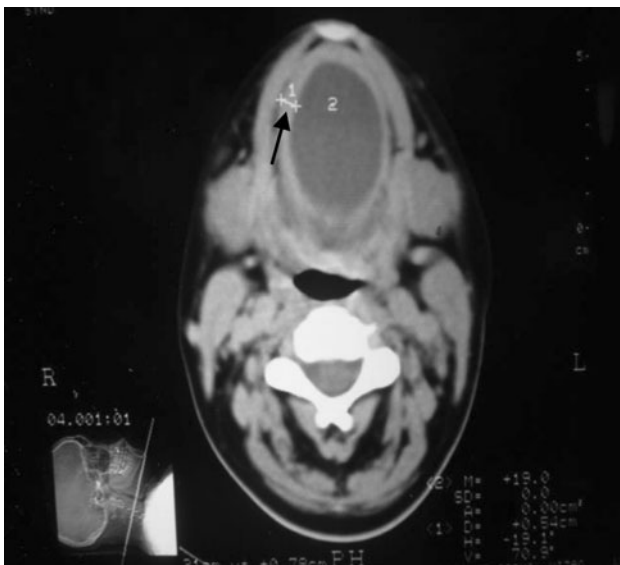


Fig. 2 Pre operative CT scan showing thick capsule but not showing much enhancement with I.V contrast and no calcifications were seen

male-to-female ratio is 1:1.3, without significant side preference [2]. Ranulas of the floor of the mouth occur in approximately 5% of patients undergoing submandibular duct relocation for the management of uncontrollable sialorrhea [3]. Presentation is most frequently in the second and third decades of life, with an age range of 3–61 years. Congenital ranulas can arise secondary to an imperforate salivary duct or ostial adhesion. These are very rare and have been known to spontaneously resolve. Post traumatic

ranulas arise from trauma to the sublingual gland, leading to mucus extravasation and formation of a pseudocyst. The more appropriate term for this may be mucous escape reaction (MER) [4]. Plunging ranulas generally appear in conjunction with an oral ranula. Rarely, they can arise independently of the oral component. Patients present first with an oral swelling in up to 45% of cases, with associated oral swelling in 34%, and without any oral involvement in 21% of cases [5].

Clinically, the oral ranula though they are generally small to medium in size, displaces the tongue, and interferes with oral function. Very large oral ranulas or ranulas located in the area of the caruncula sublingualis may lead to partial obstruction of the Wharton duct resulting in submandibular swelling during eating. In our case report obstructive symptoms were not observed.

Plunging Ranulas

Plunging ranulas arise in the neck by three mechanisms:

- The sublingual gland may project through the mylohyoid, or an ectopic sublingual gland may exist on the cervical side of the mylohyoid. This explains most plunging ranulas that exist without an oral component [6].
- The cyst may penetrate through the mylohyoid. Up to 27–45% of mylohyoid muscles in cadavers are found to be dehiscent, usually in the anterior two thirds of the muscle. These sites of dehiscence provide a route of egress for the cyst. In some instances, surgical trauma from initial ranula operations may scar or fibrose the superior surface of a ranula. When the ranula recurs, the path of least resistance is through a dehiscent mylohyoid, and a plunging ranula forms when only a simple ranula was present initially. Up to 44% of all plunging ranulas are iatrogenically induced in this manner.
- A duct from the sublingual gland may join the submandibular gland or its duct, allowing ranulas to form in continuity with the submandibular gland. Therefore, the ranula accesses the neck from behind the mylohyoid muscle [6].

The diagnosis of ranula is based principally on the clinical examination and on computerised tomographic or magnetic resonance imaging findings for the plunging ranula. When it is an isolated oral lesion, the diagnosis is generally easily accomplished. The suspicion of the plunging ranula is definitely increased if evidence of a ranula has been seen intraorally with the cervical swelling. However, when ranulas present as a cervical swelling without an oral component, the diagnosis should be properly considered. Lesions that should be considered include

Fig. 3 a, b Post operative CT scan

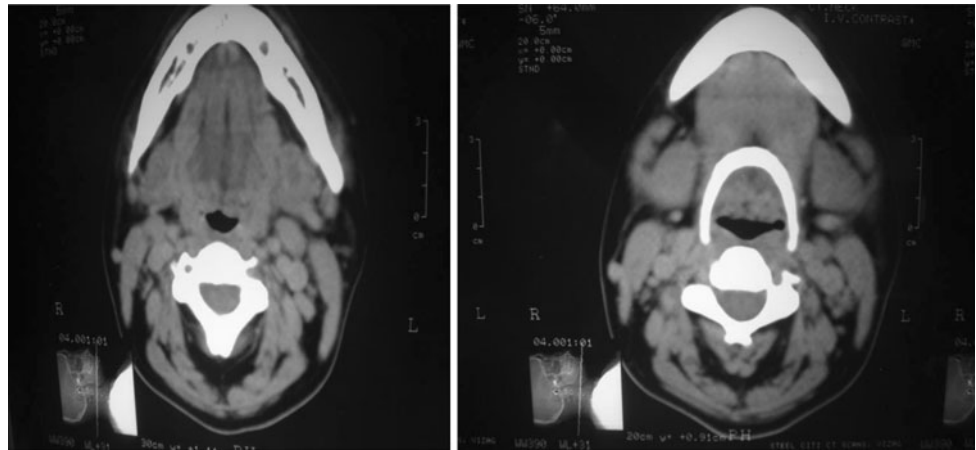


Table 1 Differential diagnosis of ranula

	Lesions	Characteristics in CT Scan	Comparision with CT scan of present case
1	Dermoid cyst and epidermoid cyst	Contains keratin and another material of high protein or fat content	Shows thick capsule and contains water
2	Lipoma	Shows low attenuation	Shows high attenuation
3	Thyroglossal cyst	Present as mid-line structure, clearly demonstrable on CT	Bilateral involvement is seen, but not present as midline.
4	Branchial cyst	Present medial to the anterior border of the sternocleidomastoid muscle	Not present medial to the anterior border of the sternocleidomastoid muscle
5	Cystic hygroma	It contains septae, not a single cavity	It is present as a single cavity
6	Ranula	Contains water, present as single cavity, bilateral involvement seen	Similar to present CT characteristic.

thyroglossal duct cysts, branchial cleft cysts, parathyroid cysts, cervical thymic cysts, dermoid cysts, cystic hygroma and benign teratoma [5]. CT scanning can provide an outline of the precise boundaries of the cyst, record the attenuation, and help narrow the range of diagnostic possibilities [4]. Dermoid cysts and epidermoid cyst contains keratin and another material of high protein and or fat content, and lipomas have low attenuation. Thus, these entities all differ in appearance in the CT scan from the plunging ranula which contains water mucin. The thyroglossal duct cyst is usually a midline structure and is usually demonstrable on the CT scan. A branchial cleft cyst is usually medial to the anterior border of the sternocleidomastoid muscle. A CT scan is especially useful in differentiating anterior cystic hygroma which generally contains septae easily identified on the CT scan and the plunging ranula which is usually a single cavity. If there is still doubt about the diagnosis, aspiration of mucous from the lesion and a laboratory determination of the amylase content should make the diagnosis of ranula obvious.

Ranulas have been managed by marsupialisation, excision of the lesion, or combined excision of both the ranula

and the gland involved [7, 8]. Treatments that do not include removal of the involved sublingual gland, such as incision and drainage, excision of the ranula, and marsupialisation, have high recurrence rates. There is no question that excision of the offending sublingual gland will cure all ranulas. The CO₂ laser [9] and ER: YAG lasers [10] is used for the excision of simple ranula which offers advantages over other modalities by sealing of blood vessels 0.5 mm in diameter or less, decreasing inflammatory response, decreasing scar contracture, eliminating suturing, and decreasing pain. Ranulas have also been managed by cryosurgery and electrosurgery but it was associated with increased tissue destruction, specimens are not available for microscopic evaluation, and excessive postoperative edema, tissue coagulation and destruction that is accompanied by increased postoperative scar contracture and pain. Recently drugs like OK-432 and Botulinum toxin type A [11] have been used for treating ranulas. OK-432 (picibanil) is a lyophilized mixture of a low virulence strain of *Streptococcus pyogenes* incubated with benzyl penicillin. The mechanism of OK-432 [8, 12] sclerotherapy of plunging ranula may be the collapse and adhesion of the

pseudocyst wall after the injected OK-432 solution is absorbed, there is extensive production of cytokine including interleukin-6 and tumor necrotic factor. These cytokines increase the endothelial permeability. The accelerated lymph drainage and increased lymph flow lead to shrinkage of the cystic spaces thereby causing an inflammatory reaction of the cystic wall rather than burning and necrosis of the epithelial cells as seen on true cysts and lymphangiomas.

Conclusion

Diagnosis and differentiation of the above presented case from other neck swellings is difficult and challenging without CT scan. Treating young patients conservatively through intra oral approach though difficult is preferred for better cosmesis.

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