Original Article | Neuroimaging and Head & Neck

http://dx.doi.org/10.3348/kjr.2016.17.2.264
pISSN 1229-6929 · eISSN 2005-8330
Korean J Radiol 2016;17(2):264-270

Korean Journal of Radiology
KJR

Plunging Ranulas Revisited: A CT Study with Emphasis on a Defect of the Mylohyoid Muscle as the Primary Route of Lesion Propagation

Ji Young Lee, MD[1, 2], Hee Young Lee, MD[1], Hyung-Jin Kim, MD[1], Han Sin Jeong, MD[3], Yi-Kyung Kim, MD[1], Jihoon Cha, MD[1], Sung Tae Kim, MD[1]

Departments of [1]Radiology and [3]Otorhinolaryngology-Head and Neck Surgery, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul 06351, Korea; [2]Department of Radiology, Hanyang University Hospital, Hanyang University College of Medicine, Seoul 04763, Korea

Objective: The purpose of this study was to clarify the pathogenesis of plunging ranulas in regard of the pathway of lesion propagation using CT scans.

Materials and Methods: We retrospectively reviewed CT scans of 41 patients with plunging ranula. We divided plunging ranulas into two types: type 1 was defined as those directly passing through a defect of the mylohyoid muscle with the presence (type 1A) or absence (type 1B) of the *tail sign* and type 2 as those through the traditional posterior route along the free edge of the mylohyoid muscle. Images were also analyzed for the extent of the lesion in respect to the spaces involved. As for type 1 lesions, we recorded the location of the defect of the mylohyoid muscle and the position of the sublingual gland in relation to the defect.

Results: CT scans demonstrated type 1 lesion in 36 (88%), including type 1A in 14 and type 1B in 22, and type 2 lesion in 5 (12%). Irrespective of the type, the submandibular space was seen to be involved in all cases either alone or in combination with one or more adjacent spaces. Of the 36 patients with type 1 lesions, the anterior one-third was the most common location of the defect of the mylohyoid muscle, seen in 22 patients. The sublingual gland partially herniated in 30 patients.

Conclusion: Our results suggest that the majority of plunging ranulas take an anterior shortcut through a defect of the mylohyoid muscle.

Index terms: *Plunging ranula; CT; Salivary glands; Anatomy*

INTRODUCTION

Ranulas are mucous extravasation cysts of the sublingual gland that have been classified as simple and plunging, with the former being confined to the sublingual space (SLS) and the latter extending beyond it (1). The diagnosis of ranulas is usually determined by the clinical features with the aid of imaging studies. Typically, they present during young adulthood, with most cases initially presenting during the third decade (1). Contrary to simple ranulas which usually present as an oral mass or swelling, plunging ranulas are often seen as a submandibular or neck mass with no clinically apparent oral connection (2). If there is doubt about the diagnosis, aspiration of mucus from the lesion for the determination of amylase content should make the diagnosis more obvious (3).

On CT and MR images, most of plunging ranulas are seen as a well-defined, unilocular, homogeneous, nonenhancing

Received May 7, 2015; accepted after revision December 31, 2015.
Corresponding author: Hyung-Jin Kim, MD, Department of Radiology, Samsung Medical Center, Sungkyunkwan University School of Medicine, 81 Irwon-ro, Gangnam-gu, Seoul 06351, Korea.
• Tel: (822) 3410-6451 • Fax: (822) 3410-2559
• E-mail: hyungkim@skku.edu

cystic mass with fluid attenuation and signal intensity, located within the submandibular space (SMS), often with contiguous involvement of the ipsilateral SLS and/or parapharyngeal space (PPS) (1, 4). Classically, communication between the sublingual and submandibular components occurs behind the posterior free edge of the mylohyoid muscle and typically appears as a smooth tapered continuation anteriorly into the SLS, the so-called *tail sign*, which is a specific sign indicating that plunging ranulas originate from the sublingual gland (1, 2, 4, 5). The *tail sign* also provides an important clue to differentiate plunging ranulas from other cystic lesions arising in or near the SMS, such as cystic hygroma, thyroglossal duct cyst, second branchial cleft cyst, abscess, and dermoid/epidermoid cyst (1, 2).

Unlike this traditional description, however, the previous studies have reported that plunging ranulas can also extend to the SMS directly through a defect of the mylohyoid muscle, instead of a classic posterior route (1, 6-10), which has not been emphasized in the radiologic literature. The purpose of this study was to clarify the pathogenesis of plunging ranulas in regard of the pathway of lesion propagation using CT scans.

MATERIALS AND METHODS

This study was approved by our Institutional Review Board, and informed consent was waived in accordance with the requirements of a retrospective study. Between January 1995 and December 2009, a search of electronic data base of our institution revealed a total of 175 patients who were diagnosed as ranula. CT scans were available for review in 78 patients. Of these 78 patients, 37 patients were excluded because the lesions were confined to the SLS (n = 17) or there was no surgical or laboratory proof of the diagnosis of plunging ranula (n = 20). Finally, the remaining 41 patients with plunging ranula formed the basis of this study and we retrospectively reviewed the CT images in these patients. There were 24 men and 17 women, ranging in age from 5 to 71 years, with a mean age of 26 years. The diagnosis was based on surgery in 23 patients and fine-needle aspiration in 18 patients. Partial or complete removal of the sublingual gland in addition to drainage of ranula was carried out with an intraoral or cervical approach. In patients who did not undergo surgery, the diagnosis was made by the analysis of mucous fluid obtained by needle aspiration to confirm the presence of amylase. Lesions were right-sided in 23 patients and left-sided in 18 patients. All patients denied a history of previous trauma or surgery in the neck.

All CT scans were obtained with a HiSpeed Advantage (GE Healthcare, Milwaukee, WI, USA) or a Somatom Plus 4 (Siemens Medical Systems, Erlangen, Germany) scanner. In all patients, postcontrast axial CT scans were obtained after the intravenous administration of 60–100 mL of iopamidol (Iopamiro 300; Bracco, Milan, Italy) or iopromide (Ultravist 300; Bayer Healthcare, Seoul, Korea) at a rate of 3 mL/s with 2.5- to 3.75-mm section thickness. Direct (n = 12) or reformatted (n = 24) coronal scans with 2- to 3-mm section thickness were also available for review in 36 patients.

CT scans were interpreted by a dedicated head and neck neuroradiologist and a general neuroradiologist in consensus, who have been practicing in the field for 23 years and 16 years, respectively. We divided plunging ranulas into two types according to the pathway through which they seemed to be formed in the SMS: type 1 was defined as those leaving the SLS anteriorly through a defect of the mylohyoid muscle (Fig. 1A-D) and type 2 as those through the traditional posterior route along the free edge of the mylohyoid muscle (Fig. 1E). The defect of the mylohyoid muscle was considered to be present, if there was a discrete discontinuation of the mylohyoid muscle or the muscle could not be followed anteriorly on CT scans.

The so-called *tail sign*, which is the core of CT diagnosis of plunging ranula, was considered to be present when there was contiguous involvement of the SLS that communicated with the SMS regardless of types of pathway (Fig. 1A, E). Often, however, there were cases where the *tail sign* was hardly recognized on CT scans. In this situation, the lesion was seen as a cyst located in the SMS, which abutted the herniated sublingual gland, without involvement of the SLS on CT scans (Fig. 1C, D). Accordingly, we further subdivided type 1 into type 1A and type 1B, depending on the presence (type 1A) (Fig. 1A, B) and absence (type 1B) (Fig. 1C, D) of the *tail sign*. Images were also analyzed for the extent of the lesion in respect to the spaces involved. As for type 1 lesions, we recorded the location of the defect of the mylohyoid muscle and the position of the sublingual gland in relation to the defect. When a part of the sublingual gland was situated in the SMS or pinched in the defect in addition to the component in the SLS, we called it partial herniation. When the entire sublingual gland was seen in the SMS without any components in the SLS, we called it total herniation.

RESULTS

CT scans demonstrated type 1 lesion in 36 (88%), including type 1A in 14 (Fig. 2) and type 1B in 22 (Fig. 3), and type 2 lesion in 5 (12%) (Fig. 4). Surgery was performed in 6 of 14 type 1A lesions, 13 of 22 type 1B lesions, and 4 of 5 type 2 lesions, respectively. Irrespective of the type, the SMS was seen to be involved in all cases either alone or in combination with one or more adjacent spaces. All 14 lesions with type 1A involved the SMS and SLS with (n = 1) or without (n = 13) involvement of the PPS. All 22 lesions with type 1B involved the SMS with (n = 7) or without (n = 15) involvement of the PPS. All 5 lesions with type 2 had a cystic component in the SLS (*tail sign*) and involved the SMS and SLS with (n = 1) or without (n = 4) involvement of the PPS.

Of 36 patients with type 1 lesions, the anterior one-third was the most common location of the defect of the mylohyoid muscle, seen in 22 patients, followed by the middle one-third (n = 8) and the combined anterior and middle thirds (n = 6). Partial and total herniation of the sublingual gland was noted in 30 patients and one patient, respectively. In the remaining five patients, the sublingual gland did not show significant displacement from the SLS.

DISCUSSION

The present study revealed that type 1 plunging ranulas

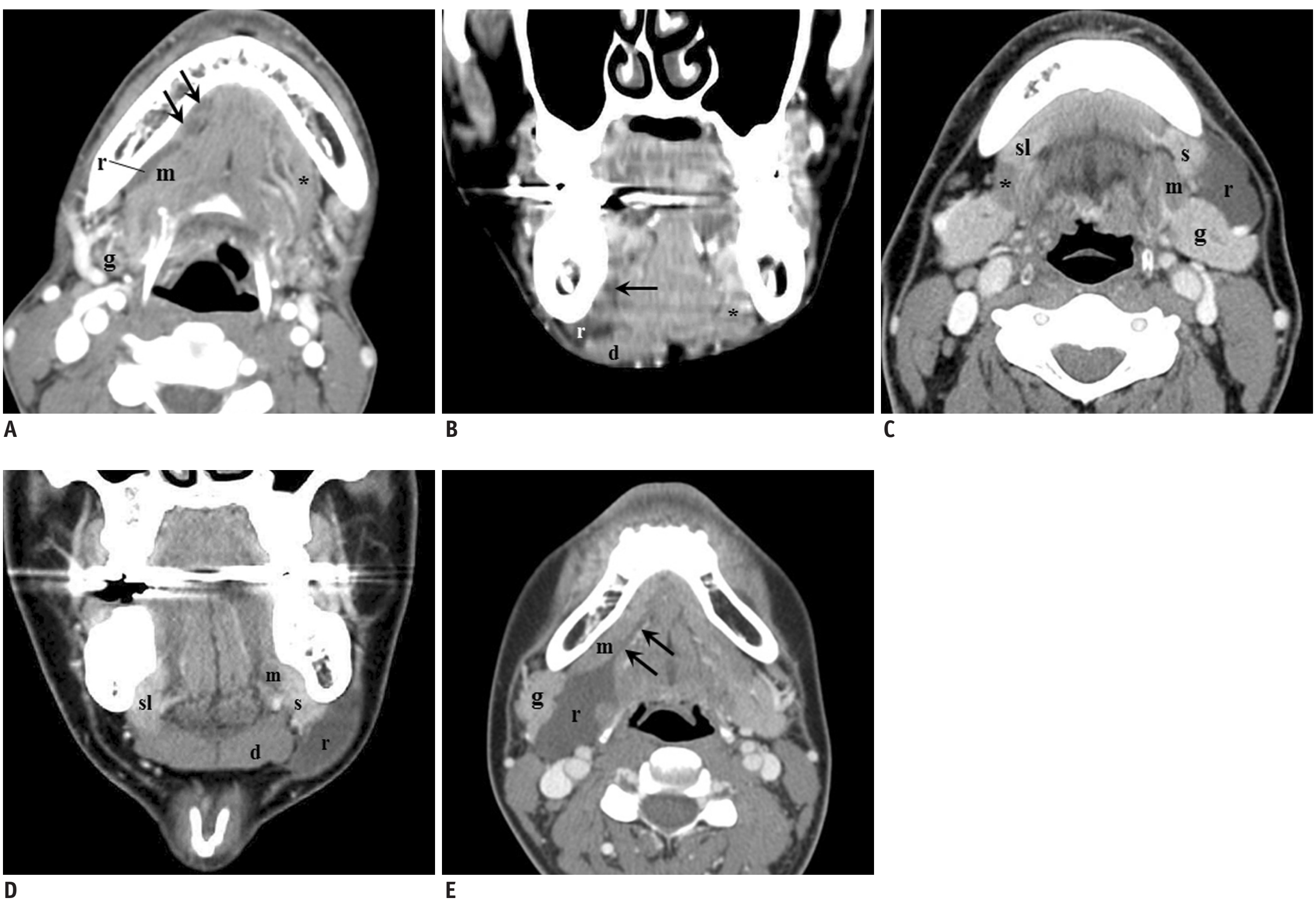
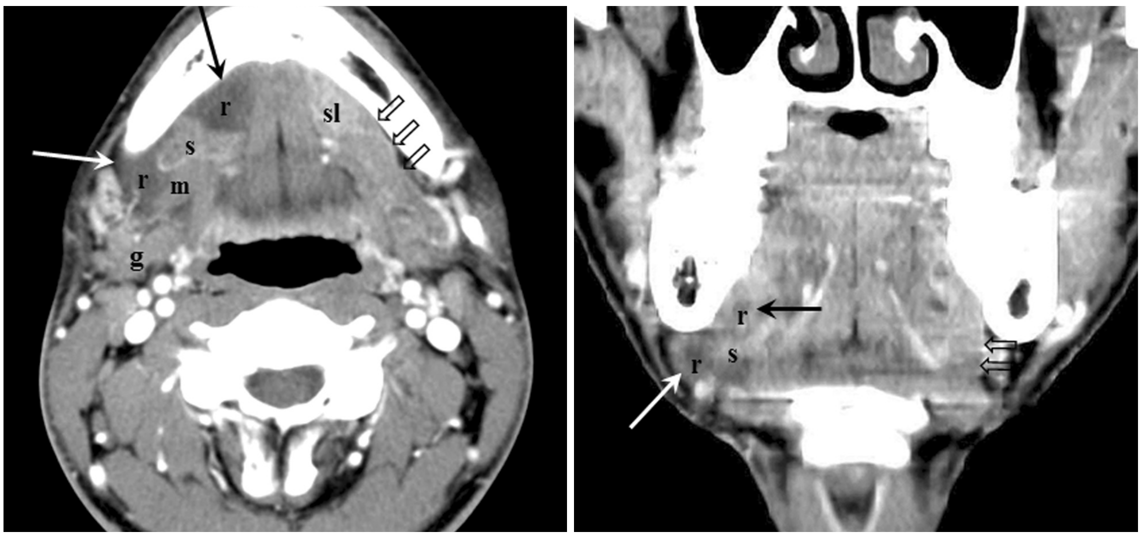


Fig. 1. Classification of plunging ranula.
A, B. Type 1A. Contrast-enhanced axial **(A)** and reformatted coronal **(B)** CT scans show elongated cystic lesion (r) in right submandibular space (SMS), lateral to mylohyoid muscle (m), which cannot be traced in anterior mouth floor. Instead, small cystic lesions (*tail sign*, arrows) are seen in ipsilateral sublingual space, contiguous to cystic lesion in SMS. Anterior part of ipsilateral mylohyoid muscle is missing on coronal image, while that on contralateral side (*) is well visualized. **C, D.** Type 1B. Contrast-enhanced axial **(C)** and reformatted coronal **(D)** CT scans show lobulated cystic lesion (r) in left SMS, lateral to mylohyoid muscle (m) and anterior to submandibular gland (g). Cyst abuts sublingual gland (s), which is displaced laterally through anterior defect of mylohyoid muscle, without evidence of *tail sign*. Also noted is even larger size of anterior defect of contralateral mylohyoid muscle (*), through which sublingual gland (sl) also herniates partially. **E.** Type 2. Contrast-enhanced axial CT scan shows large lobulated cystic lesion (r) in right SMS, posterior to mylohyoid muscle (m) and medial to submandibular gland (g). Beak with narrow channel directing to sublingual space is noted at anterior portion of cyst (*tail sign*, arrows). d = digastric muscle

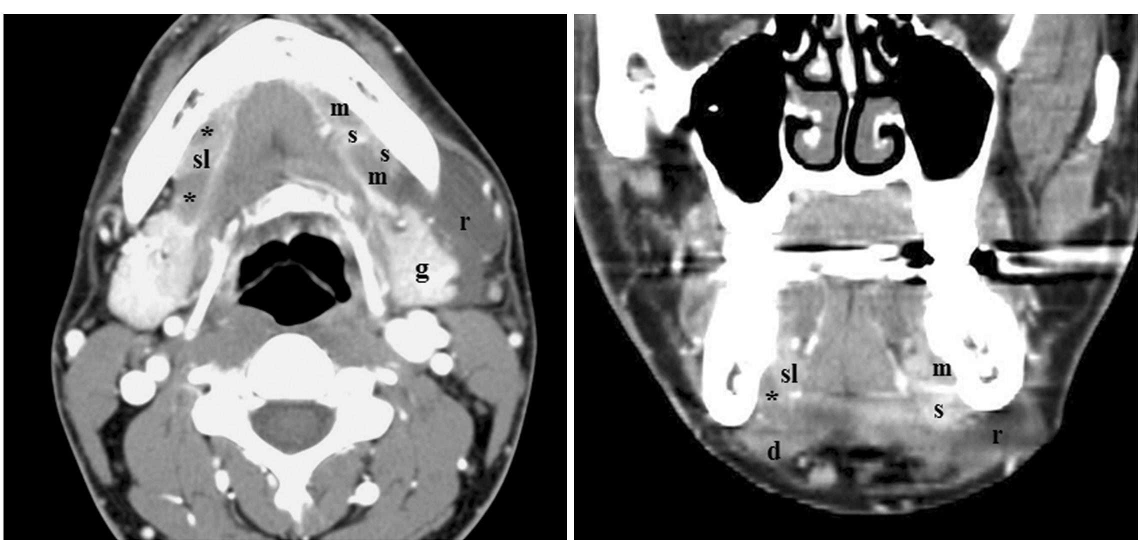
Korean Journal of Radiology KJR

were much more common than type 2 lesions (88% vs. 12%). Our results are in contrast to those reported previously (1, 2, 4, 5), which usually emphasized the classic posterior route along the free edge of the mylohyoid muscle, defined as type 2 in this study, as the major pathway of propagation of plunging ranulas. In the literature, most of plunging ranulas associated with a defect of the mylohyoid muscle, defined as type 1 in this study, have been reported as small case series. Only a few studies mostly from New Zealand have emphasized the role of mylohyoid defect as the important pathogenesis of plunging ranulas (7, 10, 11). Reportedly, plunging ranulas are prone to occur in the Maori and Pacific Island Polynesian populations (10). Although the precise etiology of their predisposition in those ethnic groups is unknown, a high prevalence of congenital defect of the mylohyoid muscle or local trauma is considered to be



A B

Fig. 2. Type 1A plunging ranula in 23-year-old man.
Contrast-enhanced axial **(A)** and reformatted coronal **(B)** CT scans show cystic lesion (r) occupying sublingual (black arrows) and submandibular (white arrows) spaces on right through large anterior defect of mylohyoid muscle (m). Sublingual gland (s) also herniates through mylohyoid defect. Within submandibular space, lesion lies anterior to submandibular gland (g). Compare mylohyoid muscle (open arrows) and sublingual gland (sl) in contralateral neck.



A B

Fig. 3. Type 1B plunging ranula in 26-year-old man.
Contrast-enhanced axial **(A)** and reformatted coronal **(B)** CT scans show large cystic lesion (r) in left submandibular space (SMS) around angle of mandible. Within SMS, lesion lies anterolateral to submandibular gland (g), contiguous anteromedially to sublingual gland (s) which herniates through defect of mylohyoid muscle (m). Note similar appearance of defect of mylohyoid muscle (*) in contralateral neck. d = digastric muscle, sl = contralateral sublingual gland

Fig. 4. Type 2 plunging ranula in 17-year-old man. Contrast-enhanced axial CT scan shows cystic lesion (r) in right submandibular space (SMS). Lesion tapers anteromedially toward sublingual space through posterior free edge of mylohyoid muscle (m), creating so-called *tail sign* (arrows). Within SMS, lesion lies posterior to submandibular gland (g).

an important factor. All patients included in our study were Far East Asian populations. It may be that ethnicity of the subjects included in different studies is really an important factor that determines the type of plunging ranulas. Further studies are recommended to illuminate the effect of ethnic variations on the pathogenesis of plunging ranulas.

Whether the ethnic variation is the critical factor in the formation of plunging ranulas or not, it is helpful to be aware of the anatomy and embryology of the mylohyoid muscle to understand the developing process of type 1 lesions. The paired mylohyoid muscle is a muscular sling that forms the floor of the mouth and separates the SLS superomedially from the SMS inferolaterally (9). It arises from the mylohyoid line on the inner aspect of the mandible, which extends from the mandibular symphysis anteriorly to the last molar tooth posteriorly. Though classically thought as a continuous muscular barrier between the SLS and SMS, the mylohyoid muscle embryologically has separate anterior and posterior parts (12). While the posterior part inserts onto the body of the hyoid bone, the anterior part inserts into the fibrous median raphe that runs from the mandibular symphysis to the hyoid bone (9, 12). The relationship between the anterior and posterior parts is such that the former is more superficial to the latter with some degree of overlap (6, 7). When the overlap is not complete, a lateral hiatus is formed between the two parts, resulting in a potential area of dehiscence through which the salivary tissue herniates (6). In his cadaveric study, Gaughran (13) referred to this anatomic relationship as mylohyoid boutonnière and sublingual bouton.

A wide variation in the incidence of the defects of the mylohyoid muscle has been reported in the literature. In the cadaveric dissection studies, the reported incidence ranged from 10% to 72% (13-16). These defects also can be detected on imaging studies. In the CT study by White et al. (12), the defects were identified in 77 (77%) of 100 asymptomatic individuals, either bilaterally in 67% or unilaterally in 33%. The authors speculated that because the detection of the far anteriorly-located defects would be limited on CT, the true incidence might be higher than reported in their study. Kiesler et al. (17) reported that 19% of European patients who had submandibular swelling due to lymphadenopathy or other masses than plunging ranula showed the defects of the mylohyoid muscle at ultrasonography. In contrast, in the study of 33 patients with plunging ranula, Jain et al. (7) found the associated defects of the mylohyoid muscle in all patients (100%) on ultrasonography.

In respect to the size of mylohyoid defects, although most defects are less than 5 mm, they may be larger than 2 cm (7, 12, 13). Defects tend to be located along the lateral margins of the mylohyoid muscle, closer to the mandible and away from the median raphe, almost always in the anterior half of the mylohyoid-mandible attachment (7, 12). Various anatomic structures pass through the defects, including salivary tissue, fat, and blood vessels (6, 12-16). According to Engel et al. (15), the histologic types of salivary tissue were either a mucous-type suggestive of sublingual gland or a mixed mucous-serous type with serous predominance suggestive of submandibular gland with a nearly equal prevalence. Blood vessels that traverse the defects are branches of submental arteries and veins arising from the facial artery and vein as well as several lymph vessels (6, 12). If the sublingual gland herniates partially, it can be located both in the SLS and SMS with a dumbbell-like appearance (9). In our study, the sublingual gland was seen to herniate partially through the anteriorly located defects of the mylohyoid muscle in most cases. In reality, however, on real-time ultrasonography the sublingual gland actively herniates through the defect of the mylohyoid muscle during tongue movement, which is seen as a tail-like protrusion at rest, as reported by Jain and Jain (11).

The clinical implications of the results of our study may not be great, because management of plunging ranulas would not be different, regardless of the types, i.e., complete or selective surgical excision of the sublingual gland with drainage of ranula via an intraoral or cervical approach to prevent recurrence. At present, most surgeons prefer the intraoral approach, because it is less destructive and leaves less scarring (3). Moreover, the location of the sublingual gland from which ranula arises may affect the length of incision at surgery. It is likely that a smaller incision is enough for the lesions anteriorly located, such as type 1 plunging ranulas, while a longer incision is needed for those posteriorly located, such as type 2 plunging ranulas. We think that the exact preoperative imaging delineation of the extent of plunging ranulas according to the types would be helpful not only to understand the pathogenesis of the disease but also to prepare the surgical planning. Recently, sclerotherapy with OK-432 or ethanol is another option to treat ranulas (18, 19).

There are two serious drawbacks in our study, which are mainly related to selection bias. First, we only included the patients whose CT scans were available for review that might have caused a serious sampling error. However, considering that most patients with plunging ranula would undergo imaging studies including CT scanning, it would not seriously affect the results of our study. Second, not all patients in this study underwent surgery. Although the diagnosis of plunging ranula is quite obvious in the patients with type 1A and type 2 lesions because of the characteristic *tail sign* on CT scans, it may be equivocal in the patients with type 1B lesions. However, in all 13 of 22 patients with type 1B lesions who underwent surgery, the surgeon could find a connection between the cyst and the sublingual gland. Absence of the *tail sign* as seen in type 2 is believed to be the additional features that support the diagnosis of type 1B plunging ranula in these patients.

In conclusion, unlike the traditional idea that plunging ranulas extend from the SLS posteriorly through the free edge of the mylohyoid muscle to enter the SMS, our results suggest that the majority take a shortcut through a defect of the mylohyoid muscle, located anteriorly near the sublingual gland. Although the exact prevalence of plunging ranulas in general population is not known, there seems to be ethnic variation between the different types of plunging ranulas.

REFERENCES

1. Macdonald AJ, Salzman KL, Harnsberger HR. Giant ranula of the neck: differentiation from cystic hygroma. *AJNR Am J Neuroradiol* 2003;24:757-761
2. Coit WE, Harnsberger HR, Osborn AG, Smoker WR, Stevens MH, Lufkin RB. Ranulas and their mimics: CT evaluation. *Radiology* 1987;163:211-216
3. 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
4. Kurabayashi T, Ida M, Yasumoto M, Ohbayashi N, Yoshino N, Tetsumura A, et al. MRI of ranulas. *Neuroradiology* 2000;42:917-922
5. Charnoff SK, Carter BL. Plunging ranula: CT diagnosis. *Radiology* 1986;158:467-468
6. Hopp E, Mortensen B, Kolbenstvedt A. Mylohyoid herniation of the sublingual gland diagnosed by magnetic resonance imaging. *Dentomaxillofac Radiol* 2004;33:351-353
7. Jain P, Jain R, Morton RP, Ahmad Z. Plunging ranulas: high-resolution ultrasound for diagnosis and surgical management. *Eur Radiol* 2010;20:1442-1449
8. Mair IW, Schewitsch I, Svendsen E, Haugeto OK. Cervical ranula. *J Laryngol Otol* 1979;93:623-628
9. Otonari-Yamamoto M, Nakajima K, Tsuji Y, Otonari T, Curtin HD, Okano T, et al. Imaging of the mylohyoid muscle: separation of submandibular and sublingual spaces. *AJR Am J Roentgenol* 2010;194:W431-W438
10. Davison MJ, Morton RP, McIvor NP. Plunging ranula: clinical observations. *Head Neck* 1998;20:63-68
11. Jain P, Jain R. Types of sublingual gland herniation observed during sonography of plunging ranulas. *J Ultrasound Med* 2014;33:1491-1497
12. White DK, Davidson HC, Harnsberger HR, Haller J, Kamya A. Accessory salivary tissue in the mylohyoid boutonnière: a clinical and radiologic pseudolesion of the oral cavity. *AJNR Am J Neuroradiol* 2001;22:406-412
13. Gaughran GR. Mylohyoid boutonni'ere and sublingual bouton. *J Anat* 1963;97:565-568
14. Castelli WA, Huelke DF, Celis A. Some basic anatomic features in paralingual space surgery. *Oral Surg Oral Med Oral Pathol* 1969;27:613-621
15. Engel JD, Harn SD, Cohen DM. Mylohyoid herniation: gross and histologic evaluation with clinical correlation. *Oral Surg Oral Med Oral Pathol* 1987;63:55-59
16. Nathan H, Luchansky E. Sublingual gland herniation through the mylohyoid muscle. *Oral Surg Oral Med Oral Pathol* 1985;59:21-23
17. Kiesler K, Gugatschka M, Friedrich G. Incidence and clinical relevance of herniation of the mylohyoid muscle with penetration of the sublingual gland. *Eur Arch Otorhinolaryngol*

2007;264:1071-1074
18. Kim JH. Ultrasound-guided sclerotherapy for benign non-thyroid cystic mass in the neck. *Ultrasonography* 2014;33:83-90
19. Rho MH, Kim DW, Kwon JS, Lee SW, Sung YS, Song YK, et al. OK-432 sclerotherapy of plunging ranula in 21 patients: it can be a substitute for surgery. *AJNR Am J Neuroradiol* 2006;27:1090-1095