Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 31 (2019) 189–191

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case Report

A case of herniation of the mylohyoid muscle with penetration of the sublingual gland

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ARTICLE INFO

Keywords:
Sublingual gland
Herniation
Mylohyoid muscle

ABSTRACT

Swelling of the submandibular region is a pathological condition that is frequently encountered in clinical practice. However, the fact that it can result from "sublingual gland herniation," in which the sublingual gland protrudes from a fissure of the mylohyoid muscle into the submandibular space, is not well known. Here, we report our experience with a case in which swelling of the submandibular region was caused by sublingual gland herniation. The patient visited our hospital with a chief complaint of swelling of the mandibular region. Detailed examination (including ultrasonography, computed tomography (CT), and functional magnetic resonance imaging (MRI) with Valsalva maneuver) confirmed the presence of a fissure in the mylohyoid muscle and protrusion of the sublingual gland into the submandibular space. As a result of no findings indicating inflammation or lymph node swelling including a malignant disease, a follow-up observation was performed. These findings suggested that it was necessary to consider the sublingual gland herniation in a case of submandibular swelling and that echographic and/or MRI examinations combined with the Valsalva maneuever were useful to distinguish.

1. Introduction

In clinical practice, cases involving submandibular swelling are frequently observed. Causes of submandibular swelling mainly include inflammatory diseases such as lymphadenitis and submandibular sialadenitis, neoplastic diseases/tumor-like diseases such as malignant lymphoma and submandibular gland tumor, cystic diseases such as sublingual ranula, and local lesions related to other systemic diseases. A precise differentiation of the cause of the swelling is therefore needed. In particular, when deep level abscesses progress, there is a risk of onset of airway compression and mediastinitis, and it is necessary to initiate treatment after making an appropriate and prompt diagnosis.

To cover many different kinds of diseases, it is important to choose the best imaging modality in consideration of cost, invasiveness and reliability. There are various methods to investigate these lesion, plain radiography, ultrasonography (US), magnetic resonance imaging (MRI), computed tomography (CT), scintigraphy and positron emission tomography (PET) [1]. Ultrasonography and magnetic resonance imaging (MRI) are used for the examination of submandibular swelling. Ultrasonography is used only in the shallow region, but it can capture images non-invasively in real time, and is therefore widely used as a simple screening technique. As with ultrasonography, MRI is also a noninvasive examination method and its constructing image are superior for evaluating the morphology of deep level tissues and the range of abscesses, because it can clearly detect water and soft tissues.

The floor of the mouth consists of the mylohyoid muscle and anterior belly of the digastric muscle. In particular, the mylohyoid muscle forms a complete barrier between the sublingual space and the submandibular space, and maintains the tissue present in the floor of the mouth in the correct position [2]. However, according to several anatomical studies on the head and neck, morphological changes such as contraction and deletion of the mylohyoid muscle have been reported [3,4]. In addition, the results of a large-scale anatomical study using the cadaver confirmed that there is a fissure in the mylohyoid muscle. According to Harrison et al. and Engel et al., a fissure was confirmed in the mylohyoid muscle in approximately 43% and 45% of cases respectively [5,6]. Therefore, the presence of a fissure in the mylohyoid muscle and the existence of a state in which the sublingual space and submandibular space communicate is not rare. To date, there are reports of the sublingual gland entering a fissure in the mylohyoid muscle and progressing to the submandibular region. Here, we report our experience with a case in which the sublingual gland progressed

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https://doi.org/10.1016/j.ajoms.2018.12.008
Received 13 October 2018; Accepted 17 December 2018
Available online 27 December 2018

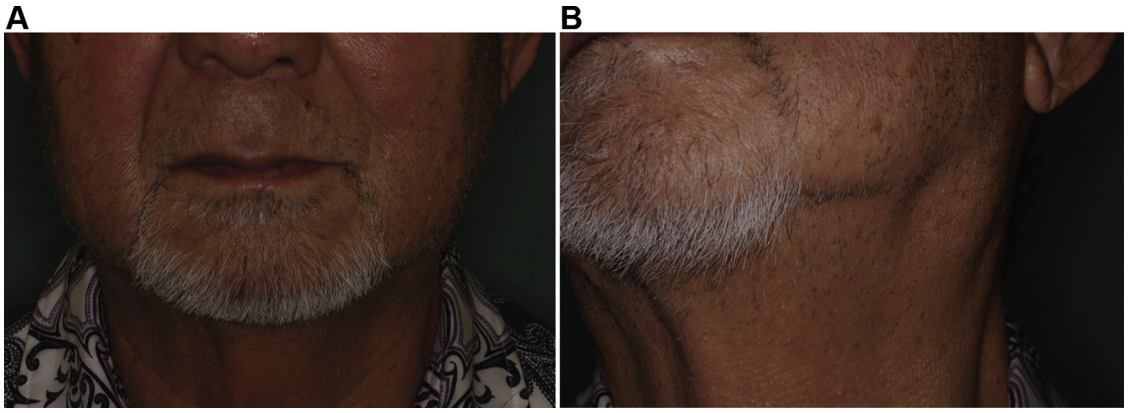

Fig. 1. Photograph of the left submandibular swelling at rest at first consultation.
(A) Frontal view: asymmetric face and normal skin color.
(B) Left lateral view: swelling of submandibular region.

into the submandibular region causing the swelling of the cervical region.

2. Case report

The patient was a 76-year-old man who presented with intermittent swelling of the left submandibular region (Fig.1A, B). Approximately three months before initial hospital visit, the patient noticed swelling in the same region, though the patient ignored it because of no symptoms without swelling. During that period, the swelling repeatedly increased and decreased in size. Physical examination revealed that an elastic soft mass with a thumb-sized head on the left mandible was palpated, without tenderness, and adhesion to the surrounding tissue. The patient was receiving outpatient treatment for diabetes, hypertension, and hyperuricemia at the department of internal medicine of the Gifu University Hospital.

In order to confirm the diagnosis, non-contrast MRI was performed with the consent of the patient, but MRI findings did not reveal the presence of lymphadenopathy or a tumor. The left sublingual gland protruded into the submandibular cavity via the defect in the mylohyoid muscle (Fig. 2). The enlarged sublingual gland was homogeneous, and had a clear margin. No imaging findings indicative of inflammation or a tumor were observed.

Neck ultrasonography was subsequently performed by a Pro-Sound α6 ultrasound system (Hitachi-Aloka Medical, Tokyo, Japan). On the B-mode examination, the presence of a mylohyoid muscle fissure was confirmed, it was observed that the left sublingual gland protruded from the fissure of the mylohyoid muscle into the submaxillary region (Fig. 3A). After explaining the Valsalva maneuver (taking a breath in and inflating the cheek) to the patient, neck ultrasonography was re-

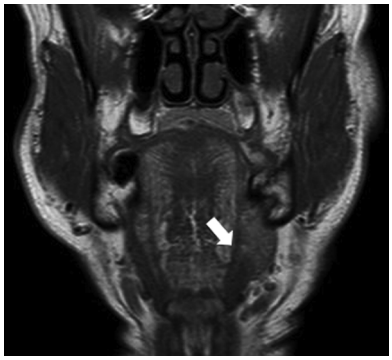

Fig. 2. Coronal T1-weighted image shows the left sublingual gland (arrow) that protrudes through the defect of the mylohyoid muscle.

performed, the herniation of the sublingual gland and the fissure in the left mylohyoid muscle became pronounced with Valsalva maneuver (Fig. 3B). As a result of these finding, mylohyoid herniation of the sublingual gland was made, no symptoms other than submandibular swelling were observed, and no enlargement in swelling was observed in the serial follow-up for several years.

3. Discussion

The floor of the mouth is formed by the mylohyoid muscle and the anterior belly of the digastric muscle. In particular, the mylohyoid muscle forms a complete barrier between the sublingual space and the submandibular space, and holds its contents in the appropriate place. When there is a defect in part of the mylohyoid muscle, the sublingual gland may protrude into the submandibular space, [3] and sublingual tissues such as fat tissues and glands may cause asymptomatic swelling in the submandibular and/or cervical region. In many cases, herniation of the sublingual tissue does not show any particular symptoms [7].

Kiesler et al. [3] classified sublingual gland herniation into four types according to the positional relationship between the sublingual gland and mandibular muscles (Table 1). Grade 0 (41%) indicates that the sublingual gland and the mandibular muscles are in their normal position. Grade 1 (40%) indicates that the sublingual gland compresses and causes thinning of the mylohyoid muscle. Grade 2 (17%) indicates the presence of a fissure in the mylohyoid muscle, but at rest, the sublingual gland does not protrude into the space. Instead, it advances from the space into the submandibular space during swallowing. Grade 3 (2%) is a state in which the sublingual gland is herniated even at rest. Such cases are rare pathological conditions. [3]

According to the classification of Kiesler et al. [3], a fissure may be present in the mylohyoid muscle at a relatively high rate. Yang HC et al. [8] reported that in their cadaveric anatomical study fissures were confirmed in the mylohyoid muscle in 29 of 50 cases, and White et al. [9] reported partial defects in the mylohyoid muscle in 77% of cases when CT scans were performed [9]. It has also been similarly reported that even in healthy adults, mylohyoid muscle fissures occur at a rate of approximately 45%–60% [10]. Based on such an anatomical viewpoint, there may be many patients in whom sublingual gland herniation develops. However, many cases of sublingual gland herniation are asymptomatic and are easily overlooked. Jain et al. [11] reported that the pathology of sublingual gland herniation might increase the susceptibility of sublingual glands to trauma and the risk of onset of sublingual ranula. Therefore, when swelling of the submandibular region is observed, it is necessary to consider the possibility of a sublingual herniation in a case.

In the literature to date, cases of submandibular swelling

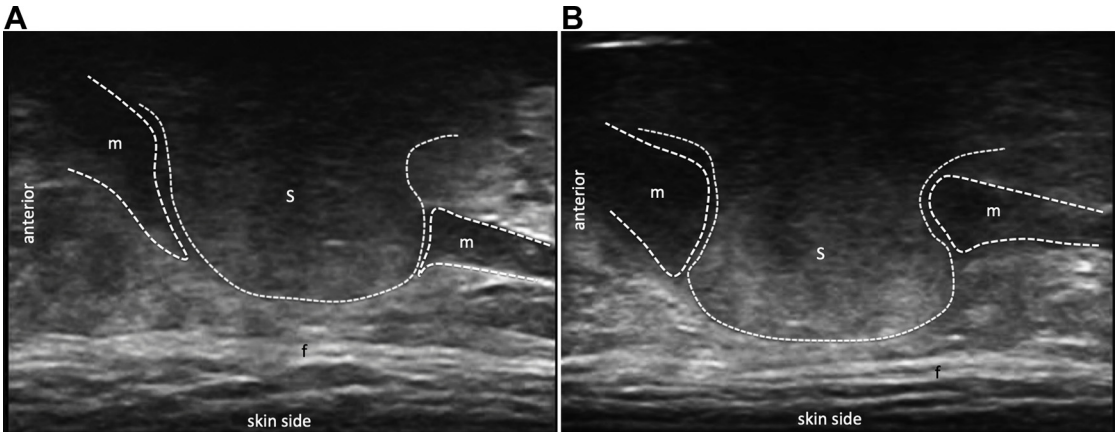

Fig. 3. Images of ultrasonography on submandibular region.
(A) The case without the *Valsalva* maneuver: there is a cleft of the mylohyoid muscle.
(B) The case with the *Valsalva* maneuver: It can be confirmed that the sublingual gland protrudes from the cleft on the mylohyoid muscle.
s: sublingual gland, m: mylohyoid muscle, f: subcutane fat.

Table 1
Classification of sublingual gland penetration by K.Kiesler et al.

Grade 0 (41%)	Normal structures
GradeI (40%)	Thinning of the mylohyoid muscle during swallowing
GradeII (17%)	Herniation of the gland during swallowing Not swell at rest
GradeIII (2%)	Herniation of the sublingual gland to cleavage of the mylohyoid muscle at rest

accompanied by sublingual gland hypertrophy as revealed by CT and MRI have been reported [12]. According to these reports, defects in the mylohyoid muscle may contain fat, blood vessels, and salivary tissue [13]. Salivary gland heterotypes can be diagnosed by a variety of examination techniques including CT, MRI, ultrasonography, salivary secretion, or nuclear medicine examination (technetium 99mTc-pertechnetate) [10,14]. Among these methods, ultrasonography is an extremely useful method as it is inexpensive (cost effectively), non-invasive (without exposing patients to radiation), accurately and quickly [11,15]. The main advantage of salivary gland ultrasonography (SGUS) is the direct visualisation of structural abnormalities of the salivary glands and surrounding tissues [16], and we can confirm ultrasonographic appearances of salivary pathologies, including infective and inflammatory conditions [17]. In particular, SGUS is usefulness for the diagnosis and the prognostic evaluation of primary Sjögren's syndrome (SS) patients [18]. So ultrasonography is the most appropriate first line investigation of shallow layer in head and neck region. In particular, ultrasound examination in combination with the Valsalva maneuver appears to be very useful for diagnosis of sublingual gland herniation. The Valsalva maneuver is a method for stimulating the vagus nerve by increasing intrathoracic pressure through inhibition of respiration, and it is used as an early treatment for paroxysmal supraventricular tachycardia (PSVT). Using the Valsalva maneuver, internal pressure increases and the sublingual gland is pushed outward so that protrusion of the sublingual gland from the mylohyoid muscle becomes prominent and easy to detect by ultrasonography.

The pathological condition in which onset of sublingual gland herniation occurs in the mylohyoid muscle fissure is currently widely unrecognized in clinical practice. However, from an anatomical viewpoint, sublingual gland herniation is observed in approximately 20% of adults [3,4]. Therefore, when examining patients who present with swelling in the submandibular and/or head and neck region, in addition to the presence of lymphadenopathy, abscesses, and submandibular gland tumors, sublingual gland herniations should be distinguished. Additionally, it is necessary to perform detailed examination with ultrasonography and MRI in combination with the Valsalva maneuver in a case.

Disclosures

None.

References

[1] Luciano N, Ferro F, Bombardieri S, Baldini C. Advances in salivary gland ultrasonography in primary Sjogren's syndrome. Clin Exp Rheumatol 2018;36(Suppl. (114)):159–64.
[2] Yang HC, Kim SY, Kim SK, Oh CS, Chung IH, Nam KI. A cadaveric study on mylohyoid herniation of the sublingual gland. Eur Arch Otorhinolaryngol 2016;273:4413–6.
[3] 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;264:1071–4.
[4] Windisch G, Weiglein AH, Kiesler K. Herniation of the mylohyoid muscle. J Craniofac Surg 2004;15:566–9.
[5] Harrison JD, Kim A, Al-Ali S, Morton RP. Postmortem investigation of mylohyoid hiatus and hernia: aetiological factors of plunging ranula. Clin Anat 2013;26:693–9.
[6] Hopp E, Mortensen B, Kolbenstvedt A. Mylohyoid herniation of the sublingual gland diagnosed by magnetic resonance imaging. Dentomaxillofac Radiol 2004;33:351–3.
[7] Keberle M, Eulert S, Relic A, Hahn D. Functional MR imaging of submandibular herniation of sublingual tissues through a gap of the mylohyoid muscle in two cases of submandibular "masses". Eur Radiol 2005;15:1326–8.
[8] Yerli H. Dynamic sonography and CT findings of unilateral submandibular gland agenesis associated with herniated hypertrophic sublingual gland. J Clin Ultrasound 2014;42:176–9.
[9] White DK, Davidson HC, Harnsberger HR, Haller J, Kamya A. Accessory salivary tissue in the mylohyoid boutonniere: a clinical and radiologic pseudolesion of the oral cavity. AJNR Am J Neuroradiol 2001;22:406–12.
[10] Ahmed M, Strauss M, Kassaie A, Shotelersuk V, DeGuzman R. Bilateral submandibular gland aplasia with clinico-radiological mass due to prolapsing sublingual salivary tissue through mylohyoid boutonniere: a case report and review. Dentomaxillofac Radiol 2009;38:121–4.
[11] Jain P, Jain R. Types of sublingual gland herniation observed during sonography of plunging ranulas. J Ultrasound Med 2014;33:1491–7.
[12] Herrera-Calvo G, Garcia-Montesinos-Perea B, Saiz-Bustillo R, Gallo-Teran J, Lastra-Garcia-Baron P. Unilateral submandibular gland aplasia with ipsilateral sublingual gland hypertrophy presenting as a neck mass. Med Oral Patol Oral Cir Bucal 2011;16:e537–40.
[13] Haktanir A. CT and MR findings of bilateral submandibular gland aplasia associated with hypertrophied symmetrical sublingual glands herniated through mylohyoid defects. Dentomaxillofac Radiol 2012;41:79–83.
[14] Schrotenboer J, Heider A, Smith E, Aronovich S. Foregut duplication cyst of the floor of mouth in a neonate: case report. Br J Oral Maxillofac Surg 2016;54:1019–21.
[15] Srinivasan A, Moyer JS, Mukherji SK. Unilateral submandibular gland aplasia associated with ipsilateral sublingual gland hypertrophy. AJNR Am J Neuroradiol 2006;27:2214–6.
[16] Bhatia KSS, Dai YL. Routine and advanced ultrasound of major salivary glands. Neuroimaging Clin N Am 2018;28:273–93.
[17] Lee KA, Lee SH, Kim HR. Diagnostic and predictive evaluation using salivary gland ultrasonography in primary Sjogren's syndrome. Clin Exp Rheumatol 2018;36(Suppl 112):165–72.
[18] Luciano N, Ferro F, Bombardieri S. Baldini C: advances in salivary gland ultrasonography in primary Sjogren's syndrome. Clin Exp Rheumatol 2018;36(Suppl 114):159–64.