



Herniation of the Sublingual Gland Through the Mylohyoid Muscle - Review Literature and Case Presentation

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Abstract

To report a case of herniation of the sublingual gland through the mylohyoid muscle in a neck dissection and a literature review about sublingual gland herniation. This condition is only superficially covered in some anatomy textbooks. Very few descriptions have been found in the anatomy literature, and it is overlooked in oral pathology. This knowledge may be of practical importance for diagnostic and surgical purposes. Recognition of mylohyoid deficiencies and sublingual glands herniation is the typical in imaging appearance of accessory salivary tissue will allow an accurate diagnosis of this benign anatomic variant and the prevention of unforeseen events during surgical procedures.

Keywords: sublingual gland, herniation, boutoniere, ranula, neck dissection, anatomical variation.

INTRODUCTION

Variations in anatomy most commonly arise in relation to the size and shape of the glands and their ductal systems. These variations have traditionally been described from the practice of cadaveric dissection; however, an increasing proportion are now being described from clinical imaging on account of the ever-improving resolution of radiological technique, such as digital subtraction sialography, MR sialography, and volumetric CT.

The mylohyoid muscle separates the lower oral cavity into submandibular space and sublingual space, in exception to do posterior margin of the oral cavity floor: the sublingual and the submandibular space [1]. The mylohyoid muscle forms the floor of the mouth, supporting the tongue and defining the boundary between the submandibular and sublingual space.

The mylohyoid muscle consists of anterior and posterior halves divided into right and left muscle bellies. The posterior portion inserts onto the body of the hyoid bone. The middle and anterior fibers insert into the fibrous median raphe that runs from the mandibular symphysis to the hyoid bone.

The sheet-like mylohyoid muscle lies superior and medially to the submandibular gland, separating the majority of it from the sublingual gland although a deep portion of the submandibular gland lies adjacent to the sublingual gland [2].

The sublingual space is a horizontally oriented, horseshoe-shaped area that sits in the oral cavity between the mylohyoid muscle inferolaterally and the genioglossus-geniohyoid complex medially. The sublingual space contains the sublingual salivary glands, the submandibular duct, and posteriorly, the deep portion of the submandibular gland.

The diaphragm of the mylohyoid muscle is reinforced in the inferior surface by the anterior bellies of the digastric muscles. Most of the standard anatomy books, as well as specialized oral surgery books, give the impression that the diaphragm of the mylohyoid muscles makes a competent floor for the mouth.

However, this concept is not true. During hundreds of dissections in anatomy laboratories, we often encounter cases in which the mylohyoid appears to have deficient fascicles.

Reviewing the original literature, forget (1870) suggested the existence of a hiatus in the mylohyoid muscle to explain the anatomical continuity between a sublingual ranula and a suprathyroid cyst [3]. This, to our investigation, is the earliest hint at the idea of a mylohyoid hiatus. Henle (1871) points out that the sublingual gland "often" (*nicht selten*) penetrates the muscle [4]. Delens (1881) made a similar observation using the term "*boutonniere musculaire*" to describe the hiatus [5]. Morestin (1897), interested in the pathology of the suprathyroid ranula, noted that frequent "prolongations" of the sublingual gland project through the mylohyoid muscle [6].

Such herniations typically occur near the submandibular or sublingual glands. These boutons were usually located in the anterior one-half or two-thirds of the anterior-posterior extent of the muscle and in the cleft between the muscle and the body of the mandible [7]. A division of the mylohyoid by a vein has also been reported [8] and occasionally by the stylohyoid muscle or Wharton's duct [9].

It has been observed a deficiency or hiatus in the mylohyoid muscle in cadavers between 27.3% to 58% [8, 10, 11]. It was bilateral between 11.3% to 26% of the cadavers [10, 11]. Between 30% to 32% cadavers, it was unilateral [10, 11]. From 63% to 100% of the deficiencies were located in the anterior third [10, 11]. More frequently on its lateral side (closer to the bone than to the median raphe) [10].

On the other hand, White *et al.*, observed by sonography that the mylohyoid defects were identified in 77% of the cases. The deficiencies were bilateral in 67% and unilateral in 33%. Accessory salivary tissue was identified in 37% of the cases. Fat and blood vessels were commonly identified within the mylohyoid defects. Sixty-one percent of the defects contained only fat. Thirty-five percent of the defects contained blood vessels [12].

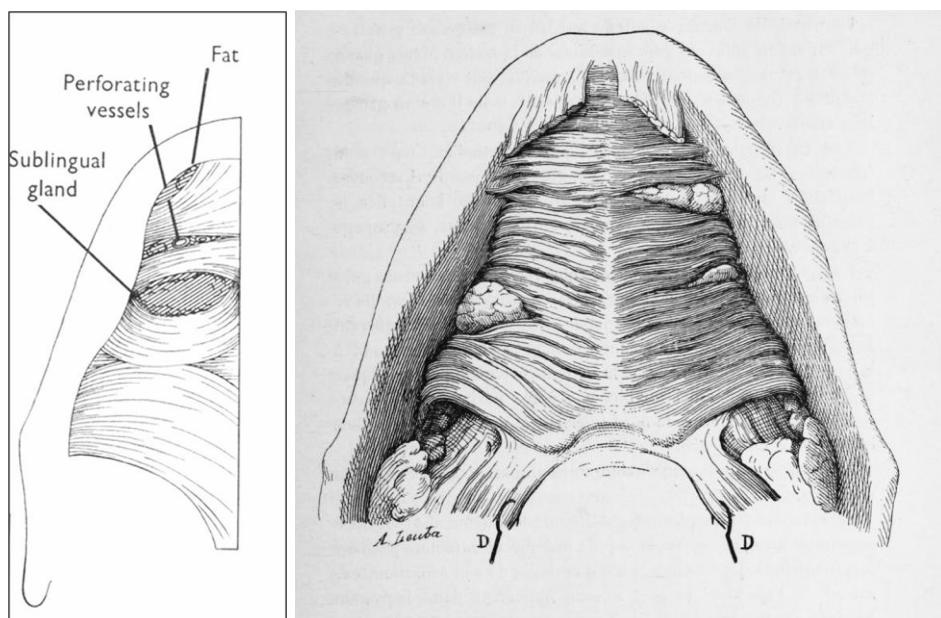


Figure 1. Right: this line drawing illustrates the superficial anatomy of the submandibular space with mylohyoid defects containing accessory salivary tissue and perforating submental branches of the facial vein and artery) [7]. Left: Inferior view of mylohyoid muscle showing sublingual glands projecting into the suprathyroid region) [6].

They appeared in variable shapes-as narrow or fusiform fissures between the transversely directed fibers of the muscle or as broader round or oval openings. Their size was also very variable-from a few millimeters to about 15 mm in maximal diameter. In several cases the hiatus was filled only with fatty tissue and covered with the mylohyoid muscle fascia, but with no bulging. In most of the cases, however, part of the sublingual gland accompanied by fatty tissue protruded and appeared bulging through the muscular deficiency [12].

As reported by Engel *et al.*, these penetrating vessels do not appear to predispose the development of accessory salivary tissue, but may be seen passing through defects with or without associated salivary tissue, or penetrating the mylohyoid muscle with no detectable defects [13].

A herniated gland may be mistaken for a soft tissue tumor or swelling [14]. It can also be mistaken as a pathological condition, both radiologically and clinically. It could cause problems when making a diagnosis and require further exploration [15], such as soft tissue tumor [14], submandibular lymph node, salivary stones of the submandibular and sublingual salivary glands, and inflammatory processes.

Morestin believed the cause of the plunging ranula, may be explained as a tear passing through the mylohyoid muscle causing the pieces of the sublingual gland to herniate [6].

In addition, some authors have reported a correlation between a sublingual gland's herniation and a plunging ranula [16, 17]. However, the evidence is ambiguous.

CASE PRESENTATION

This is a 65-year-old female patient with a history of adenocarcinoma of the palate, T3N0M0, Stage III. Clinical examination of the neck reveals a tumor at level Ia of the neck. A hemimaxillectomy, elective supraomohyoid neck dissection and temporal muscle flap was planned.

Under general anaesthesia, the neck was approached via a Kocher incision. After raising the skin and platysma flap, as well as the inferior flap. We began the dissection of level Ia of the neck in the interdigastic area.

As the dissection progressed to level Ib from the digastric tendon upwards to remove the submandibular gland, we observed glandular tissue on the same plane as the superficial part of the submaxillary gland and bluntly dissected it.

We were initially confused upon noticing the mylohyoid muscle behind this glandular tissue, so we continued blunt dissection until the submandibular gland was identified and retained in the specimen to be removed.

The neck dissection was continued at the other levels until completed. The patient progressed uneventfully and was discharged from the hospital on the third day.

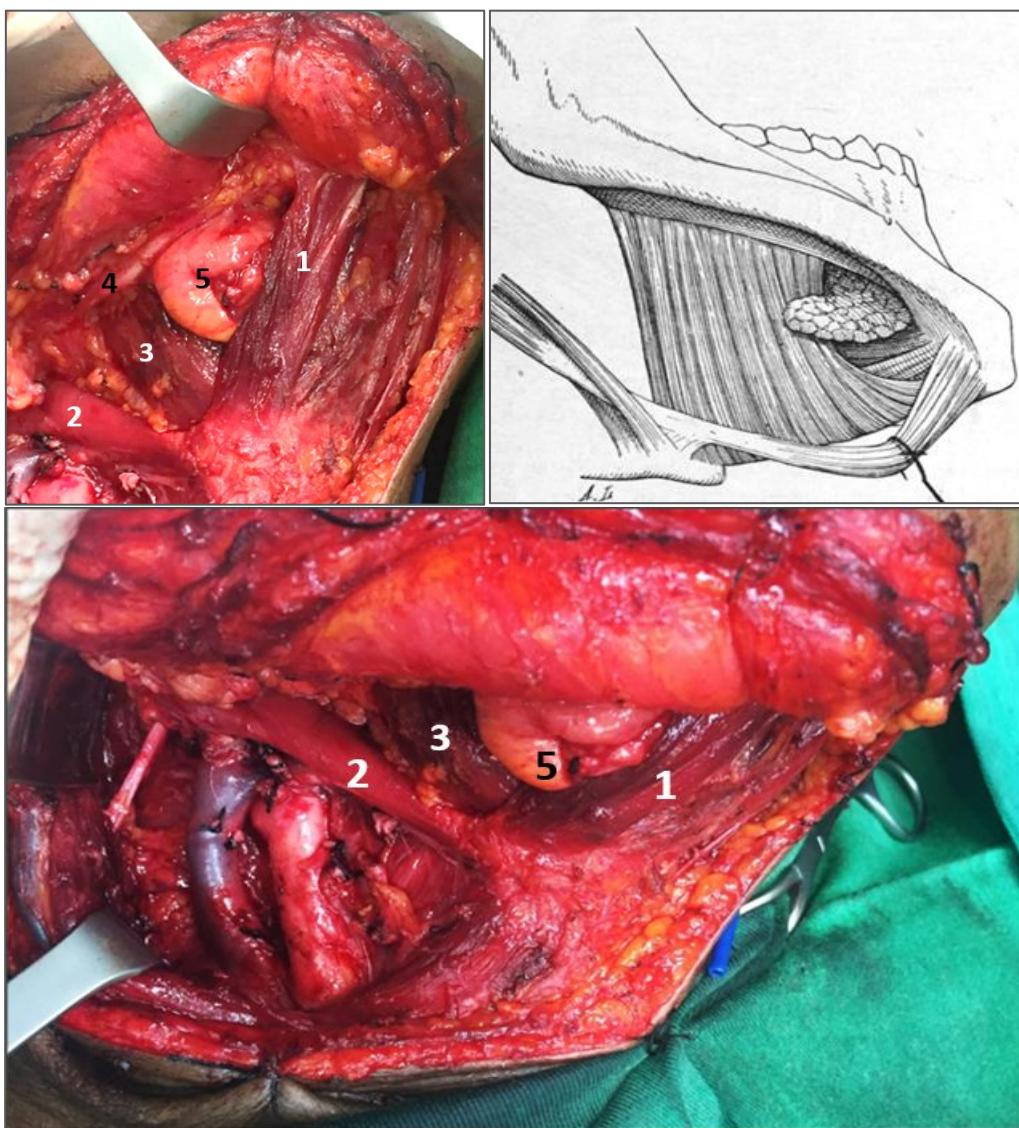


Figure 2. Transsurgical view of the herniated sublingual gland behind anterior venter of digastric muscle and in the middle of the mylohyoid muscle. The submandibular gland was removed as part of the supraomohyoid neck

dissection. 1- anterior venter of digastric muscle; 2- posterior venter of digastric muscle; 3- mylohyoid muscle; 4- mandible; 5- submandibular gland.

DISCUSSION

The presence of a mylohyoid buttonhole with a sublingual herniation represents an anatomical variation that deserves more than casual interest in anatomists and surgeons. From Hippocrates to the last century, a multitude of treatments have been applied, with surgery being the one that has prevailed when understanding the etiology of ranulas [18].

The mylohyoid muscle is not a strict anatomical barrier [7, 10, 19, 20]. Anatomical hiatus exist in the mylohyoid diaphragm through which portions of the sublingual salivary gland or fat may protrude [7, 20]. Is not well known, when do the sublingual gland protrude from a fissure of the mylohyoid muscle into the submandibular space.

It is difficult to establish whether the deficiency of the muscle and the herniation of the gland is a congenital condition or if it is developed during life [10].

In surgery involving the removal of either of these glands it is important for the surgeon to be aware of the possible existence of a sublingual gland herniation located in the submandibular region.

Herniation was classified in cadaveric study as either marginal or penetrated types sublingual gland herniated through the margin of the mandible and the mylohyoid muscle in the marginal type herniation. sublingual gland herniated by a penetrating mylohyoid muscle in the penetrated type of herniation [11].

Kiesler *et al.* [20] classified sublingual gland herniation by ultrasonography into four types according to the positional relationship between the sublingual gland and mandibular muscles (Table 1). Grade 0 (41%), Grade 1 (40%), Grade 2 (17%), Grade 3 (2%) is a state in which the sublingual gland is herniated even at rest. Such cases are rare pathological conditions [3].

TABLE. 1: Ultrasonography Classification of Sublingual Gland Penetration

Grade 0	Normal anatomical structures, no thinning of mylohyoid muscle, no signs for glandular penetration
Grade I	Circumscribed thinning of the mylohyoid muscle, vertical reduction in muscle thickness during swallowing, no signs for glandular penetration
Grade II	Herniation of the gland during swallowing, normal anatomical structures during rest
Grade III	Constant hiatus in the mylohyoid muscle with permanent herniation of the sublingual gland

The frequency of occurrence and possible clinical significance of this condition deserves its recognition in our teaching and anatomical and surgical texts [7]. 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) [21, 22]. Among these methods, ultrasonography is extremely useful as it is inexpensive (cost effectively), non-invasive (without exposing patients to radiation), accurately and quick [16, 23].

However, from an anatomical viewpoint, sublingual gland herniation is observed in approximately 20% of adults [15, 20].

Herniations through the mylohyoid muscle are not only of anatomic interest, but may be of importance in oral and maxillofacial surgery for differential diagnosis of swelling in the region, as of lymph nodes, cysts, and tumors, and for the selection of an appropriate surgical procedure [9].

These portions of the sublingual gland protruding into the hiatus of the mylohyoid muscle may allow cervical mucous extravasation [24].

The deficiencies in the mylohyoid may play a role in the spread of infections through the various compartments of the region [25], and can cause confusion when dissecting the level I in the neck. Ectopic sublingual glands below this muscle have also been reported [19, 26].

Many authors stated that meticulous extra-oral dissection of the cervical ramification of the plunging ranula in continuity with the sublingual salivary gland or even the submandibular gland is essential [27-33].

Conclusion

To report a case of herniation of the sublingual gland through the mylohyoid muscle, observed during the intraoperative period of a neck dissection.

Mylohyoid muscle defects are common. Accessory salivary tissue is frequently identified in association with mylohyoid muscle deficiencies. Occasionally, large deposits of accessory salivary tissue are seen.

Herniations through the mylohyoid are not only of anatomic interest but may be of importance in oral and maxillofacial surgery for differential diagnosis of swelling in the area such as of lymph nodes, cysts, tumors, and to select the most appropriate surgical procedure.

The deficiencies in the mylohyoid muscle may play some role in the spread of infections through the various compartments of the region [25].

As such, sublingual glands herniation should be considered when a submental neck mass is evaluated. Recognition of mylohyoid deficiencies and the typical imaging appearance of accessory salivary tissue will allow an accurate diagnosis of this benign anatomic variant and the prevention of unforeseen events during surgical procedures.

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