Bilateral Ectopic Submandibular Glands in the Carotid Triangles: A Case Report with Review of Literature

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ABSTRACT

This case report presents a rare and unusual bilateral ectopic location of the submandibular glands in the carotid triangles of the infrahyoid region of the neck. Both submandibular glands were found anteromedial to the carotid sheath suspended from the floor of the mouth by their ducts and from the lingual nerves by branches to the glands. Such rare variations are crucial in diagnostic imaging, aesthetic surgery, radiation therapy, and surgical treatment of diseases comprising those of the glands. The consideration of such unusual location is of relevance in the differential diagnosis of neck mass and the decision-making process regarding approaches to the submandibular glands during various surgical procedures (trans-cervical, trans-oral, endoscopic, etc.) and in avoiding the risk of injury to adjacent neurovascular structures such as facial vessels, cervical and mandibular branches of the facial, hypoglossal, and lingual nerves.

Keywords: Ectopic; Submandibular gland; Submandibular duct; Lingual nerve; Carotid triangle

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INTRODUCTION

The three pairs of major salivary glands (SGs) (parotid, submandibular, and sublingual) and several hundreds of minor salivary glands secrete the saliva in the mouth a major part of which is released by the submandibular glands (SMGs) alone [1]. The anatomical location of the SMG in the submandibular or digastric triangle (SMT) and its relationship to the surrounding structure is well described by previous investigators [2]. Each SMG consists of a larger superficial lobe and a smaller deep lobe, which are continuous around the posterior border of the mylohyoid muscle (MS) [2-5]. Deep to the skin, both SMGs are related to various tissue layers and are crossed over by blood vessels and nerves [2,3]. The secretion of the SMG is drained by submandibular or Wharton duct (SMD) that opens into the floor of the mouth at the sublingual papillae (caruncle) on both sides of the lingual frenulum [4-6].

During development, the secretory portions of all SGs arise from the epithelium of the oral mucosa, while their stroma is derived from the neural crest [7,8]. Their further morphogenesis is dependent on the cell-cell interactions that involves a wide range of signaling pathways [9-11] and their cells and acini mature in the last two months of gestation and continue to increase in size up to two years after birth [12]. It is well known that different variations of the SGs occur during this developmental process and ectopic salivary glandular tissue has been widely reported at several sites in the head and neck region to as far down as the rectal mucosa [13-19]. The unilaterally or bilaterally absence (aplasia) of SMG [20-22] and the presence of heterotopic glandular tissue such the thyroid gland tissue in the SMG are also well documented [23].

The saliva secreted by SGs plays multiple roles that include mechanical cleansing of food and microorganisms, lubrication of the mucosa, protection of the teeth and mucosa by its antimicrobial activity, dissolution of taste compounds, facilitation of speech, mastication, swallowing, initial digestion of starch and lipids, and esophageal clearance and gastric acid buffering [24]. Moreover, evidence shows that, in addition to saliva, the SGs secrete endocrine hormones, growth factors, and biomarkers and play essential roles in both innate and adaptive immunity, and protection [25,26]. Additionally, saliva as a body fluid is used clinically for non-invasive diagnostic testing and monitoring of various infectious and non-infectious diseases, and for the detection of illicit drug use [27-32].

As demonstrated by several functional and imaging studies, ptosis (descent or drooping) of the SMGs [33,34] and functional changes such as xerostomia occur during aging [35-38]. It was also noted that, with increasing age, there is a loss of functional glandular parenchyma with increasing amount of fibrous tissue, fat and oncocytes [39-43].

Other clinical studies show that the SMGs are prone to infectious and non-infectious inflammatory diseases, neoplastic and non-neoplastic diseases, and mechanical ductal obstructions by salivary calculi (sialolithiases) that affect their normal functions [44,45]. Clinically, these glands can be targeted during surgical procedures in the neck region for diseases in which the SMGs are not directly involved, such as SMG transfer surgery during radiation therapy and SMG-mobilization, or SMG resection for neck rejuvenation (neck lift) that can distort their location and impair their function. Regarding diseases of the SMGs, surgical excision of the glands remains the treatment of choice for malignancies as well as for refractory diseases leading to impaired glandular functions [46-50]. Moreover, diseases of structures in the neck, particularly those found in the carotid triangle and carotid space, may manifest with space-occupying lesions (mass) that can be mistaken for diseases of the SMG [51].

This case report presents a rare bilateral ectopic location of the SMGs in the carotid triangles suspended from the floor of the mouth and the lingual nerve by SMDs and a branch of the lingual nerves to the glands. Therefore, awareness of variations such as this is important in the differential diagnosis and diagnosis of neck mass, therapeutic management of diseases in the neck region including those of the glands and in avoiding iatrogenic injuries to the gland and associated structures.

MATERIAL AND METHODS

During observation of the neck of a 77-year-old male donor, an unusual bilateral submandibular and upper neck fullness was encountered. Dissection of the neck, removal of the skin, the superficial fascia with the platysma muscle, and the investing fascia revealed oval glandular masses in the carotid triangles (CTs). Then the CTs and digastric triangles (DGTs) were carefully dissected and cleaned to look for the SMGs, the SMDs, and associated nerves including the hypoglossal and lingual nerves. Photographs were taken and the nerves and ducts were painted yellow and green respectively using Microsoft paint for illustration.

RESULTS

During exposure of the neck for dissection, it was observed that the upper portion of the neck was enlarged with a barely visible inferior margin of the mandible. Removal of the skin, the superficial fascia with the platysma, and the investing fascia revealed bilateral oval glandular masses in the CT with superior and inferior poles (Figure 1). Further dissection and cleaning of these areas showed no glandular tissue in the SMTs but only glandular ducts and lingual nerves (LNs) with their branch to the glandular masses, suspending the masses from the floor of the mouth and the LNs respectively. The ducts originated from the medial aspects (deep surfaces) of the superior poles and the lingual nerve branches to the glandular masses also entered the same region of the superior poles (Figure 1 and 2). The glandular masses and the ducts were then determined to be ectopically located SMGs with no superficial and deep parts and SMDs draining into the oral cavity. The superior and inferior poles of SMGs partly overlapped the posterior belly of DGM and the stylohyoid muscle (SHM) superiorly and the superior belly of omohyoid muscle (OHM) inferiorly (Figure 1 and 2). The SMDs and the branches of the LNs to the glands crossed over the posterior belly of DGMs and SHMs just posterolateral to the body of the hyoid bone. The SMDs and the LNs with their branches were located in the SMTs below the lower margin of the mandible. After the removal of the body of the mandible and the mylohyoid muscles (MHMs), a deeper dissection of the SMTs and part of the floor of the mouth revealed that the SMDs undercrossed the LNs deep to the MHMs. Both the nerves and the ducts coursed further on the inferior surface of the hyoglossus muscle (HGM) before diving deep into the floor of the mouth (Figure 2).

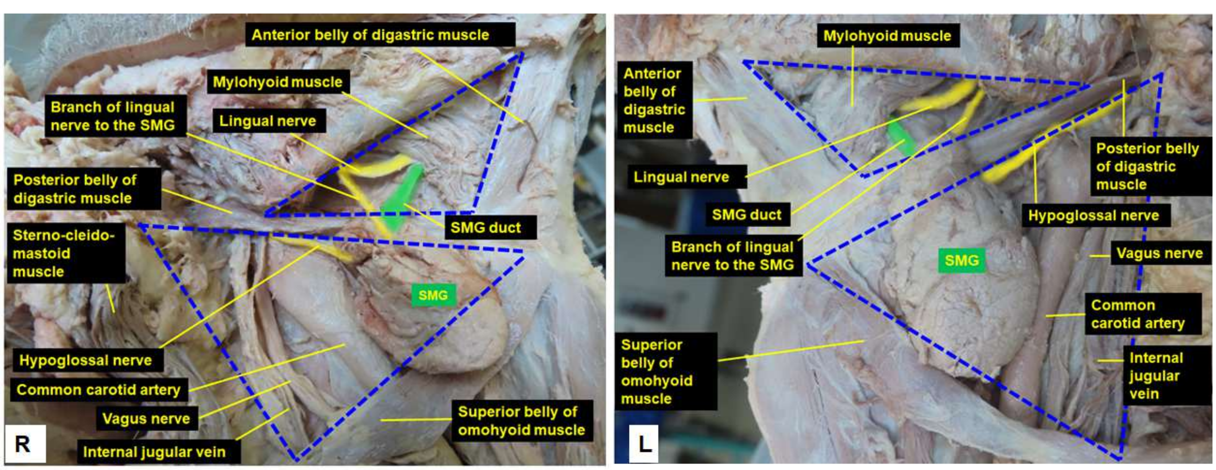

Figure 1: Illustrates the bilateral ectopic SMGs in the CT and the SMDs ascending in the SMTs to accompany the LNs to extend to the floor of the mouth deep to the MHM. The ducts and the nerves are unusually low in position and visible inferior to the lower margin of the mandible.

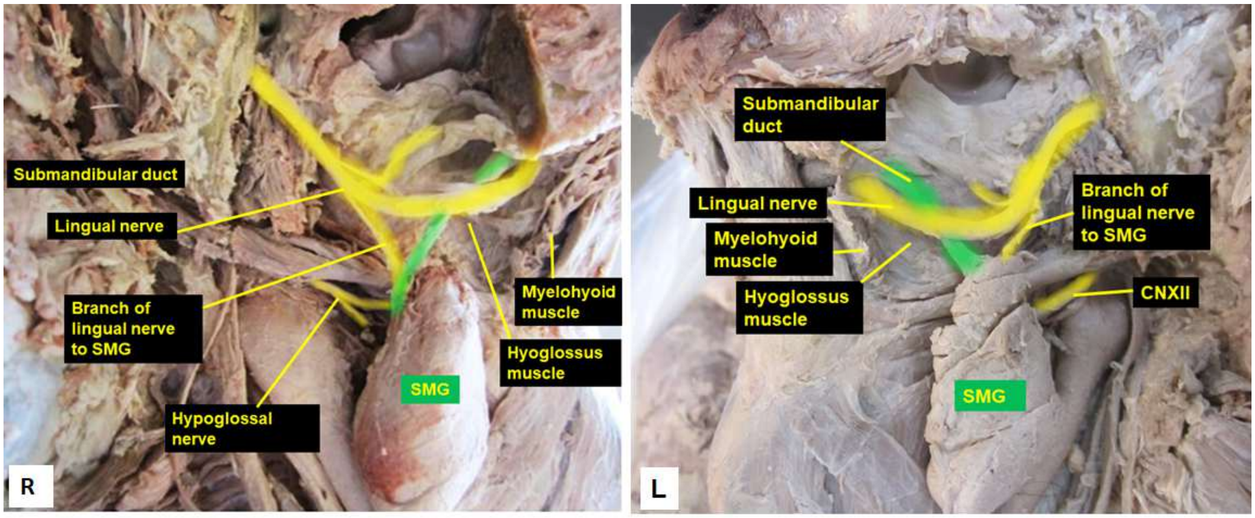

Figure 2: Deep dissection of the right and left SMT and part of the floor of the mouth after removal of the body of the mandible and the MHMs to show the courses and relationships between the SMDs and the LNs.

DISCUSSION

Out of the 500-1500 ml of saliva secreted per day, 90% is released by the three pairs of major SGs, 70 - 75% of this is secreted by the seromucous SMGs alone [1]. The SMGs are normally located in the SMT of the suprahyoid region of the neck and in the floor of the mouth. Each SMG consists of larger superficial and smaller deep lobes, which are continuous around the posterior border of the MHM [2]. The superficial lobe is covered by layers of tissues that include the skin, subcutaneous fat with the platysma muscle, the investing fascia and is crossed over by fascial vessels and the cervical branch of facial nerve. The deeper surface of the superficial lobe lies on the MHM, LN, nerve to mylohyoid, submental vessels and hypoglossal nerve (HGN) [2,3]. However, according to Eaton KJ et al. 2019 [3], the neurovascular structures that relate to the deep surface can sometimes pierce through the SMG and can be considered aberrant structures. The superficial lobe of the gland extends anteriorly to the anterior belly of DGM, posteriorly to the stylomandibular ligament, superiorly to the medial surface of the body of the mandible, and inferiorly it overlaps the intermediate tendon of DGM and the insertion of the SHM [4,5]. The deep lobe of the gland lies in the sublingual space between the MHM inferolaterally, the hyoglossus (HGM) and SHM medially, LN superiorly and the hypoglossal nerve (HGN) inferiorly [4,5]. However, the finding in the current report reveals that both SMGs have no distinct superficial and deep lobes and were bilaterally located in the CTs being suspended from the floor of the mouth by their SMDs, and from the LNs by branches to the glands. Specifically, they were located inferior to the SHM and posterior belly of DGM, anteromedial to the carotid sheath and superior to the superior belly of OHM. The superior and inferior poles of each gland were found to partly overlap the posterior belly of DGM and the SHM superiorly, and the superior belly of OHM inferiorly. The HGNs ran deep to the superior aspect of the glands before crossing under the intermediate tendon of DGM and the SHM to pierce the floor of the mouth.

The about 5 cm long SMD arises by the confluence of multiple tributaries on the medial aspect of the SMG just at the level of the posterior border of MHM. It courses between the MHM and HGM and then between the sublingual gland and genioglossus muscle. It opens into the floor of the mouth at the sublingual papillae (caruncle) on either side of the lingual frenulum. As each duct runs on the HGM, it passes between the LN and HGN, and near the anterior border of the HGM it is crossed over by the LN [5,6]. However, many studies have shown variable crossing patterns between the SMD, LN, and HGN [6]. In the current report, though the SMD arose from the deep or medial aspect of the glands near their superior poles, they originated in the CT, crossed over the posterior belly of DGM and SHM with the LN branches to the glands. Then they under crossed the LN in the sublingual space before piercing through the HGM. This course and pattern of crossover of the SMD and the LN affirms to previous findings but no close relationship with the HGN nerve was observed.

Age-related ptosis and complete absence of the SMGs are also well documented. According to the CT imaging investigations conducted on hundred consecutive by Lee et al. [33], the SMGs undergo age-related average descent (ptosis) of 0.17mm/year as measured by the distance of the bottom of the gland from the plane of the inferior border of the mandible and concluded that there is a linear relationship between age and SMG ptosis regardless of sex. However, a subsequent MRI study on 129 adult patients (mean age 52.3 years) found a significantly larger inframandibular volume with no change in the total volume and height of the SMG, which provided evidence of SMG ptosis as a significant contributor of age-related submandibular fullness particularly in males and overweight obese individuals [34]. This finding contradicts with the result of Lee et al. [33] above that showed SMG ptosis occurs with age regardless of sex. Age related functional changes of the salivary glands causing dryness of the mouth (xerostomia) are also common complaints among the elderly [35-37], which is a subjective sign of dry mouth that may or may not be accompanied by objective signs of hyposalivation [38]. Moreover, other previous functional studies of the SMG did note that with increasing age, there is a loss of functional glandular parenchyma (reduction in the number of acini) with increasing fibrous tissue, fat [39-42] and oncocytes that can be found in benign and malignant conditions [43] causing decreased salivary secretion with age for which reason about 25% of elderlies suffer from age-related Xerostomia and related complaints.

In addition to their biomedical relevance, the variations of the SMGs are clinically important for different reasons: i) They are involved in various viral, bacterial, and fungal infections, and ductal obstructions hindering salivary drainage. About 80% of salivary calculi (sialolithiases) are related to SMGs [44,45]. ii) They are affected by various non-infectious inflammatory diseases, autoimmune diseases as well as by non-neoplastic and neoplastic diseases [44,45]. iii) They may be affected by surgery in the neck region for diseases in which the SMG is not involved, such as submandibular gland transfer surgery to facilitate gland shielding during radiation therapy and SMG-mobilization, resuspension/ plication or submandibular resection during rhytidectomy for neck rejuvenation (neck lift) [46,47]. iv) Their surgical excision remains the treatment of choice for tumors and other treatment refractory diseases [48,49], which can be performed either trans-cervically or trans-orally [50]. v) They might mimic diseases of structures found in the carotid triangle and carotid space that may manifest with space occupying lesions (mass) such as lymphoma, paraganglioma, schwannoma, lipoma, carotid body tumor, localized neurofibroma, carotid sheath meningioma, carotid artery dissection, aneurysm and pseudoaneurysm, etc. [51].

Despite the extensive effort made to find whether a similar variation has ever been documented in literature, no results similar to this current case report could be found and therefore, as to the knowledge of the author, this report of bilateral ectopic SMGs in the carotid triangles suspended from the floor of the mouth by the SMDs and from the lingual nerves by branches to the glands is the first of its kind.

CONCLUSION

Such unusual bilateral ectopic location of the SMGs in the carotid triangles is noteworthy in the differential diagnosis of neck mass in diagnostic imaging, aesthetic surgery, radiation therapy, and in the surgical treatment of diseases in the neck region including those of glands such as Ludwi Angina, where the decompression of submandibular space may be needed. Therefore, awareness of such unusual location of SMGs can be helpful in the decision-making process regarding the surgical approaches to the region, particularly the SMGs, during various procedures (transcervical, transoral endoscopic, etc.) and in avoiding the risk of iatrogenic injury of associated structures such as facial blood vessels, mandibular and cervical branches of the facial nerve, lingual nerve, and hypoglossal nerve.

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