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CLINICAL RECORD

Submandibular gland ectopia associated with atrophy of floor of mouth muscles

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Abstract

Objective: We describe a rare case of an ectopic submandibular gland associated with atrophy of the ipsilateral floor of the mouth muscles.

Method: Case report and review of the world literature regarding ectopic submandibular glands.

Results: The reported patient had an ectopic submandibular gland associated with atrophy of the ipsilateral anterior digastric and mylohyoid muscles. This implies maldevelopment of these muscles in the floor of the mouth and arrest of the normal migration of the submandibular gland. The condition was diagnosed using magnetic resonance imaging and conventional submandibular gland sialography.

Conclusion: Submandibular gland ectopia in the floor of the mouth is a rare phenomenon. The described case represents the first report of an ectopic submandibular gland associated with atrophy of the ipsilateral floor of the mouth muscles. Radiologists and clinicians should familiarise themselves with this entity and its imaging findings, in order to prevent unnecessary biopsy of this benign condition.

Key words: Submandibular Gland; Pathological Conditions; Anatomical

Introduction

Submandibular gland ectopia in the floor of the mouth is an extremely rare phenomenon and has been described only once previously.[1]

To the best of our knowledge, the presented case represents the first report of an ectopic submandibular gland associated with atrophy of the ipsilateral anterior digastric and mylohyoid muscles. This implies maldevelopment of these muscles and arrest of the normal migration of the submandibular gland.

Case report

A 39-year-old man was referred in June 2008 to the otolaryngology department of Northwick Park Hospital by his general practitioner. The patient complained of intermittent prandial pain and swelling in the left parotid and sublingual regions for the last six months.

Clinical examination revealed slight swelling in the floor of the mouth and left parotid region, with no tenderness or palpable stones. The parotid and submandibular duct ostia were normally positioned.

Ultrasound examination showed normal parotid glands and a single, level three reactive lymph node. Access to the submandibular glands was limited by the patient's extensive beard.

Magnetic resonance imaging (MRI) of the neck revealed that the left submandibular gland was absent from its expected position. However, a mass was seen in the left sublingual space, associated with atrophy of the left anterior digastric and mylohyoid muscles (Figure 1). This mass resembled the normal right submandibular and sublingual salivary glandular tissue on all sequences, raising the likelihood that it represented either an ectopic submandibular gland or a hypertrophic sublingual gland (in the absence of a left-sided submandibular gland).

For further evaluation, conventional sialography of the left submandibular gland was arranged. This revealed a short salivary duct and the presence of a large salivary gland with normal intraglandular architecture (Figure 2), consistent with a normal ectopic submandibular gland lying in the floor of the mouth.

The patient was informed of the findings, reassured and discharged.

Discussion

During week six of embryological development, the submandibular gland develops from the endoderm in the floor of the mouth and migrates laterally to its definitive site in the submandibular region. Numerous branches from deep within the gland coalesce to form the main submandibular (Wharton's) duct.[2]

The paired submandibular glands consist of both superficial and deep lobes. The larger superficial lobe is located in the submandibular space, posterior and inferior to the mylohyoid muscle. The smaller superior portion (deep lobe) extends superiorly over the posterior margin of the mylohyoid muscle as a finger-like projection (the uncinate process) to enter the sublingual space. Wharton's duct courses from the deep lobe, over the mylohyoid muscle in the sublingual space along the floor of the muscle, to open

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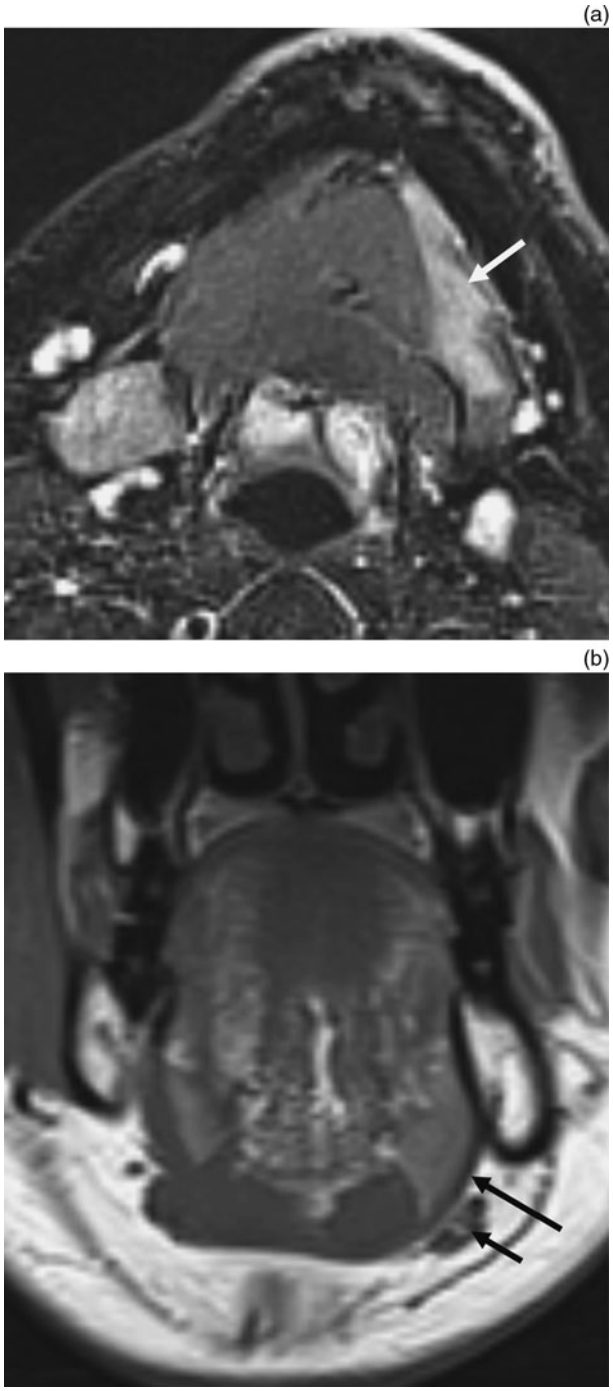

FIG. 1

(a) Axial and (b) coronal magnetic resonance imaging scans showing a mass of similar signal intensity to the right submandibular gland in the left sublingual space (white arrow), in keeping with an ectopic submandibular gland. Note the atrophied mylohyoid muscle (long black arrow) and anterior digastric muscle (short black arrow) on the left. The contralateral submandibular gland is unremarkable.

near the midline in the sublingual papilla on the ipsilateral side of the frenulum of the tongue.[3–5]

The sublingual glands are situated in the submucosa of the floor of the mouth and are bounded inferiorly by the mylohyoid muscle. The sublingual gland lacks a single dominant duct. Instead, it is drained by approximately 10 small ducts (the ducts of Rivinus) which exit the superior aspect of the

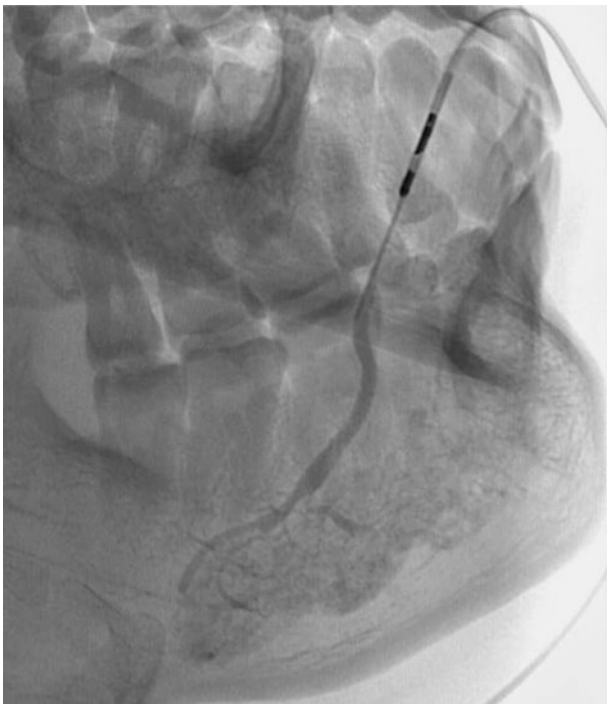

FIG. 2

Left submandibular gland sialogram confirming the ectopic position of the left submandibular gland in the floor of the mouth. Normal ductal architecture is demonstrated.

gland and open along the sublingual fold on the floor of the mouth. Occasionally, several of the more anterior ducts may join to form a common duct (Bartholin's duct), which typically empties into Wharton's duct.

In our patient, imaging revealed salivary tissue representing an ectopic submandibular gland in the floor of the mouth rather than in the gland's expected position. Ectopic salivary tissue is rare, and most authors agree that anomalous embryological development of salivary tissue is the main cause.[1,6] It is likely that, in our patient, the submandibular gland arrested along its migration pathway and came to lie in the floor of the mouth. This is the first case report describing an ectopic submandibular gland associated with atrophy of the ipsilateral anterior digastric and mylohyoid muscles. It implicates maldevelopment of these muscles in the arrest of normal submandibular gland migration.

A PubMed search was undertaken using the following key words: ectopic salivary gland, absent salivary gland, heterotopia and heterotopic salivary gland. Submandibular gland ectopia in the floor of the mouth is a rare phenomenon and has been described only once previously.[1] Ectopic salivary gland tissue has been reported to occur in numerous sites within the head and neck region, including the lateral and posterior neck, tongue, middle ear, thyroid, pituitary gland, and mandible.[7–14]

Congenital absence of the salivary glands is infrequent and more often involves multiple major salivary glands. This entity can accompany other developmental anomalies, such as mandibulo-facial dysostosis (Treacher-Collins syndrome),[15] atresia of the lacrimal puncta[16] and congenital malformations of the temporomandibular component.[17] Our patient had no other associated anomalies, as described above.

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Our patient's MRI findings could have been interpreted as showing an absent submandibular gland with compensatory ipsilateral hypertrophy of the sublingual gland, since the sublingual gland was not seen separately to the prominent salivary tissue present in the floor of the mouth. However, the prominent mass of salivary tissue in the floor of the mouth was subsequently confirmed by sialography to represent an ectopic submandibular gland rather than a hypertrophied sublingual gland. Sialography demonstrated that the glandular tissue seen in the floor of the mouth was draining through the normal submandibular duct orifice.

- **This is the first case report describing an ectopic submandibular gland associated with atrophy of the ipsilateral anterior digastric and mylohyoid muscles**
- **Clinical and radiological features are presented**
- **Radiologists and clinicians should familiarise themselves with this entity, in order to prevent unnecessary biopsy of this benign condition**

Isolated unilateral submandibular gland aplasia associated with ipsilateral sublingual gland hypertrophy has however been reported in the literature.[18] In this case report by Srinivasan *et al.*, the diagnosis of compensatory ipsilateral sublingual hypertrophy was made by computed tomography and MRI alone. Here, sialography could have been performed to confirm the underdevelopment of Warthin's duct or the absence of an ectopic submandibular gland.

Some authors believe that compensatory enlargement of a salivary gland is not feasible, as the absence of one gland would be unlikely to significantly affect the total amount of saliva produced provided that the remaining major and minor salivary glands were functioning normally.[19]

Radiologists and clinicians should familiarise themselves with the clinical picture and radiological findings for an ectopic submandibular gland associated with atrophy of the ipsilateral floor of the mouth muscles, in order to prevent unnecessary biopsy of this benign condition.

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