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## A case report of isolated agenesis of bilateral submandibular glands with compensatory hypertrophy (SIALOSIS) of other salivary glands

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### Abstract

The congenital absence of major salivary glands can be diagnosed through various imaging techniques such as ultrasound, CT, MRI, salivary ductography or radioisotope scanning. Sialosis or sialadenosis is characterized by non-inflammatory, benign, enlargement of the salivary glands due to acinar hypertrophy. It can also happen as compensatory phenomenon secondary to maldevelopment of other glands. We report a rare case of bilateral parotid hypertrophy (sialosis) associated with bilateral submandibular gland agenesis, demonstrated via ultrasound and MRI. The precise imaging modality like ultrasound or MRI obviates the need for intervention procedures to achieve the correct diagnosis.

**Keywords:** Bilateral submandibular gland agenesis, sialosis, sialadenosis

### Introduction

The congenital absence of the submandibular gland (SMG) is an uncommon condition, with around 40 cases documented in medical literature so far. However, only a limited number of case reports highlight the significance of using different imaging techniques for diagnosis [1]. The term “aplasia” can be complete or partial, isolated or diffuse and can involve the parotid, submandibular and sublingual glands. It may result primarily from cessation of organogenesis or from glandular atrophy secondary to denervation, use of adrenergic antagonists, duct ligation, obstruction [3]. Some patients are asymptomatic, while others report difficulty in chewing and swallowing due to dry mouth or tooth decay.

### Case History

A 40 year old male with a painless swelling over bilateral pre-auricular regions which was gradually increasing in size over the span of 20 years. He did not suffer from dryness of mouth or eyes. On physical examination, non-fluctuant, soft swelling over bilateral preauricular regions was palpated without rise in local temperature or redness. Intraoral examination revealed soft, non-fluctuant swelling over bilateral sublingual region. A differential diagnosis of autoimmune or neoplastic aetiology was considered.

The patient underwent a comprehensive panel of blood tests, including complete blood count, C-reactive protein, rheumatoid factor, antinuclear antibody and anti-cyclic citrullinated peptide. These tests could not conclusively concur for autoimmune or infective etiology.

A non-ultrasound guided FNAC of bilateral parotid glands was conducted which revealed many clusters of benign acinar cells hypertrophy with bare nuclei and granular cytoplasm. Subsequently, ultrasonography was performed to exclude a neoplastic cause, revealing diffusely enlarged bilateral parotid and sublingual glands (Figure 1) with a heterogeneous echotexture and normal vascularity. Bilateral submandibular glands were not visualized, but several discrete, non-necrotic lymph nodes were observed in bilateral level Ib areas of neck. A computed tomography (CT) scan was performed, which ruled out chronic sialolithiasis as the cause of submandibular gland atrophy.

Magnetic Resonance Imaging (MRI) was performed to confirm the absence of bilateral submandibular glands. The MRI showed enlarged bilateral parotid (Figure 2), sublingual, lingual and minor salivary glands (Figure 3) at the bilateral tonsillo-lingual sulci, posterior one third of the tongue, and soft palate submucosal region, all demonstrating normal

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enhancement and diffusion restriction, with corresponding low ADC values. An accessory left parotid gland was noted along the left Stenson's duct (Figure 3).

### Discussion

Major salivary glands usually originate from the proliferation and evagination of the oral ectoderm during the fourth to eighth weeks of fetal development. Agenesis of the salivary glands occurs as a result of defects during this developmental phase [1]. It occurs in about 1 in 5,000 births [1] and was first described by Gruber in 1885 as bilateral submandibular gland agenesis [2].

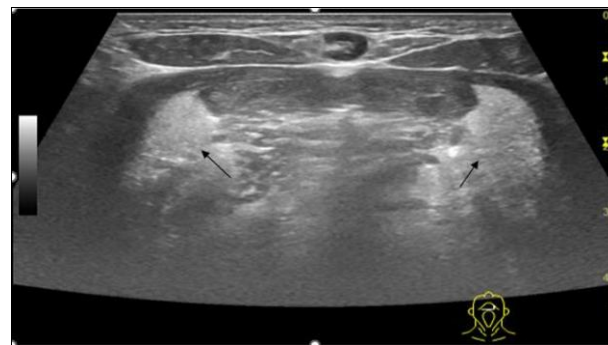
SMG agenesis is often detected through imaging. A bimanual examination of the neck alone is not adequate to establish a diagnosis. Several imaging techniques are employed, such as MRI, sialography, CT scans, ultrasound, and nuclear medicine methods, including technetium-99m pertechnetate scintigraphy. Ultrasonography is typically the initial test but can be challenging to interpret. Scintigraphy, however, is limited to revealing the presence of functionally active tissue. For a comprehensive diagnosis, cross-sectional imaging methods such as MRI or CT are recommended, as they offer better delineation of the anatomy.

A similar case reported by S.A. Kelly *et al.* [5] described a 28-year-old male with unilateral agenesis of the parotid gland, accompanied by sialosis in the contralateral parotid gland where CT sialography demonstrated a normal ductal system, diffuse enlargement of the left parotid gland and absence of the right parotid gland. To evaluate parotid function and rule out agenesis of other salivary glands, a Tc99m pertechnetate scintigraphy was conducted, which corroborated the CT findings.

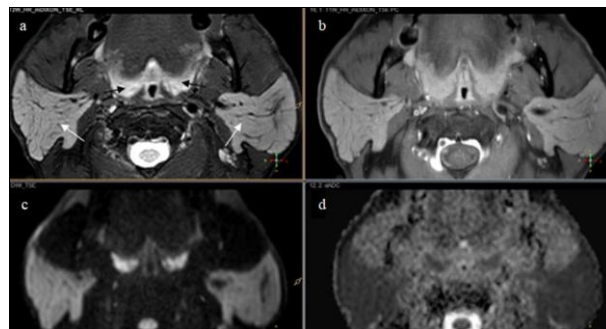
Common diseases that cause bilateral parotid gland hypertrophy include Sjögren's syndrome, sarcoidosis, various infiltrative pathologies like amyloidosis or hemosiderosis, and HIV-associated salivary gland disease [4]. However, the current case did not exhibit any symptoms consistent with these conditions. Diagnosing sialosis requires ruling out inflammatory conditions, especially sarcoidosis, Sjögren's syndrome, lymphoepithelial diseases, and HIV infection. While sialography can be helpful in diagnosis, it is rarely indicated. A distinctive 'leafless winter tree' appearance may be observed, where the absence of arborization results from the disruption of secretory ducts and the compression of smaller interlobular canaliculi [4]. MRI is the preferred imaging modality for investigating salivary gland conditions. Biopsy or fine needle aspiration cytology may conclude diagnosis but are seldom required in cases of bilateral symmetrical involvement.

Common causes of sialosis include diabetes, hypothyroidism, acromegaly, uraemia, alcoholism, certain medications such as antihypertensives and malnutrition. In our case, submandibular gland agenesis resulted in compensatory hypertrophy of the bilateral parotid glands, left accessory parotid, sublingual, lingual, and other minor salivary glands leading to sialosis.

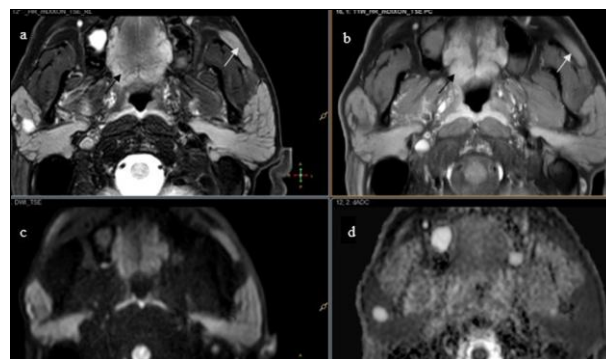
Awareness about these imaging findings is crucial for diagnosis and treatment planning. Without this knowledge, bilateral parotid swelling could be misdiagnosed as an autoimmune disorder or neoplastic condition, potentially leading to unnecessary invasive diagnostic procedures like FNAC or biopsy.



**Fig 1:** Ultrasonography of submental region (submentovertical projection) reveals diffusely bulky bilateral sublingual glands with heterogenous echotexture (black arrows).



**Fig 2:** On axial T2W DIXON (a), axial T1W post contrast (b), axial DWI (c) & ADC (d): Bilateral parotid (white arrows) and minor salivary glands (black arrows) appear bulky with multiple T2 hypointense septae showing diffusion restriction with corresponding low ADC.



**Fig 3:** On axial T2W DIXON (a), T1W post contrast (b), axial DWI (c) & ADC (d): Minor salivary glands along soft palate (black arrow) and Accessory left parotid gland (white arrow) along the left Stenson's duct.

### Summary and Conclusion

- We strongly recommend that clinicians familiarize themselves with submandibular gland (SMG) agenesis and its imaging findings.
- The absence of the SMG can lead to compensatory sialosis in the remaining salivary glands.
- Imaging techniques such as MRI and ultrasound are useful for diagnosis. Recognizing this condition is crucial to avoid misdiagnosis, additional invasive investigation and ensure appropriate treatment.

### Conflict of Interest

Not available.

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Not available.

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