

Unilateral Submandibular Gland Agenesis with Aberrant Ductal Termination Presenting as Chronic Intraoral Salivary Fistula with Xerostomia and Dental Caries: A Rare Case Report with Embryological insight.

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Abstract

Background: Submandibular gland agenesis is a rare congenital anomaly, typically asymptomatic due to compensatory salivary mechanisms. However, when associated with ductal abnormalities, it may present with atypical clinical features.

Case Presentation: A 54-year-old female presented with chronic salty discharge from the posterior floor of the mouth for 2–3 years, associated with xerostomia and dental caries. Examination revealed micrognathia, a palpable left submandibular gland, and non-palpable right gland. Intraoral examination showed a fistulous opening near the left lower molar region. MRI sialography confirmed absence of the right submandibular gland and abrupt truncation of the left submandibular duct.

Conclusion: This case highlights a rare combination of unilateral gland agenesis with contralateral ductal anomaly presenting as salivary fistula. Associated xerostomia, dental caries, and micrognathia suggest a broader developmental correlation.

Keywords: Submandibular gland agenesis, salivary fistula, xerostomia, dental caries, MRI sialography, micrognathia

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Introduction

Congenital salivary gland agenesis is an uncommon developmental anomaly, most frequently affecting the parotid glands. Submandibular gland agenesis is significantly rarer and is often detected incidentally due to compensatory salivary secretion from remaining glands [1].

However, reduced salivary flow may lead to xerostomia, increased dental caries, and oral discomfort [2]. Ductal

anomalies such as ectopic openings or truncations are even more uncommon and rarely reported in association with gland agenesis.

We present a rare case of unilateral submandibular gland agenesis with contralateral ductal truncation presenting as chronic intraoral salivary fistula, along with xerostomia, dental caries, and micrognathia—suggesting a possible embryological association



Case Presentation

A 54-year-old female presented to the ENT outpatient department with:



Persistent salty fluid discharge from the posterior floor of the mouth

- Left-sided localization near posterior tongue
- Duration: 2–3 years
- Associated xerostomia
- History of dental caries

There was no history of pain, swelling, fever, dysphagia, or trauma.

Radiological Findings: MRI Sialography\

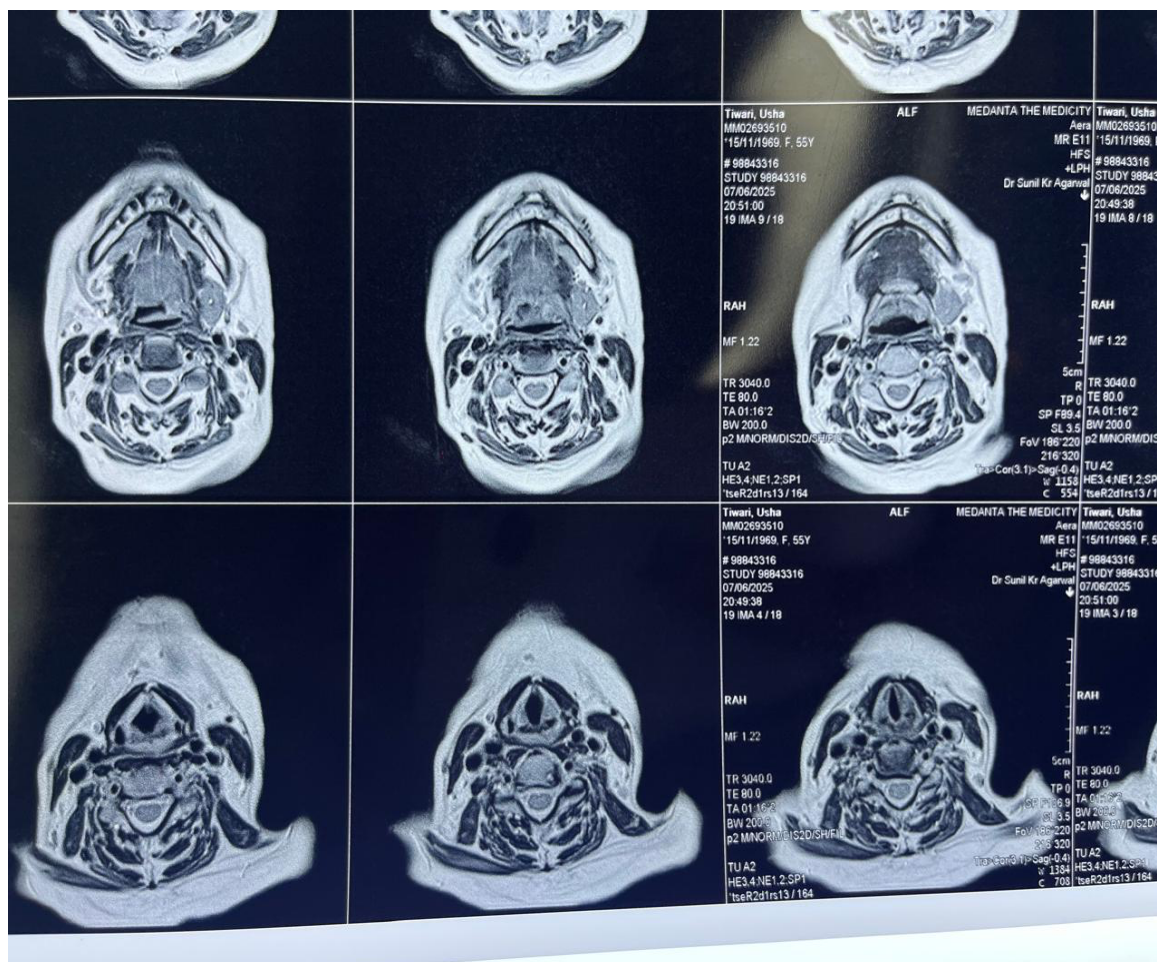
Clinical Examination

- Facial profile: Micrognathia
- Neckexamination:
Left submandibular gland: palpable, mildly enlarged
- Right submandibular gland: not palpable

Intraoral Findings

- Fistulous opening near left lower last molar region
- No mucosal lesion or ma

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Dental implant artefacts partially obscured visualization

- No mass lesion in oral cavity

Key findings:

- Right submandibular gland: not visualized → agenesis
- Left gland: normal morphology
- No inflammation or mass

Ductal Findings

- Mild duct prominence (~1.3 mm)
- Abrupt termination ~1 cm from hilum
- Distal duct not visualized
- No sialolithiasis

Additional Findings

- Small benign cervical lymph nodes
- Deviated nasal septum
- Normal parotid glands

Discussion

Submandibular gland agenesis results from failure of epithelial bud formation during embryogenesis of the first branchial arch derivatives [3].

Xerostomia and Dental Caries

Saliva plays a vital role in:

- Lubrication
- Antibacterial defense
- Enamel protection

Reduced salivary flow leads to:

- Xerostomia
- Dental caries
- Increased oral morbidity

This correlates strongly with findings in our patient [2,4].

Micrognathia Association

Although not commonly reported in isolated cases:

- Both mandible and submandibular gland arise from first arch structures
- Developmental disturbances may affect both simultaneously

Literature shows associations in syndromes such as:

- Treacher Collins syndrome
- First arch developmental anomalies [5]

In our case, isolated micrognathia suggests a possible embryological link, making this case noteworthy.

Unique Features

- Unilateral submandibular gland agenesis
- Contralateral ductal truncation
- Chronic intraoral salivary fistula
- Xerostomia + dental caries
- Associated micrognathia

Differential Diagnosis

- Salivary fistula (traumatic/infective)
- Sialolithiasis
- Minor salivary gland fistula
- Branchial arch anomalies

Imaging ruled out all secondary causes.

Management

Treatment options include:

- Conservative management
- Surgical fistula excision
- Duct reconstruction

In This Case

The patient was counseled regarding all options.

Due to mild symptoms, she opted for conservative management, and is under regular follow-up.

Conclusion

This case represents a rare and clinically significant entity combining:

- Submandibular gland agenesis
- Ductal anomaly
- Salivary fistula
- Xerostomia and dental caries
- Possible embryological link with micrognathia

Early recognition and imaging are essential for accurate diagnosis and management.

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