Alqalam Journal of Medical and Applied Sciences. 2025;8(4):2750-2752
https://doi.org/10.54361/ajmas.258494

Case report

Imaging-Based Diagnosis of an Ectopic Submandibular Gland: A Rare Clinical Presentation

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Abstract
Ectopic submandibular gland (SMG) is a rare developmental anomaly that can mimic other neck masses, leading to diagnostic challenges. We presented a rare case with a right-sided ectopic SMG. A 52-year-old female presented with a 1-year history of chronic mouth dryness (xerostomia) and intermittent dysphagia. Ultrasound revealed an abnormal position of the SMG; contrast-enhanced magnetic resonance imaging (MRI) confirmed a right-sided ectopic SMG located lateral and superior to the thyroid lobe with dilatation of Wharton's duct and features suggestive of acalculous sialadenitis. A minor salivary gland biopsy performed to exclude Sjögren's syndrome showed mild lymphocytic infiltration without autoimmune features. As no obstructive or autoimmune pathology was identified, the patient was managed conservatively with symptomatic treatment and scheduled for regular follow-up. An ectopic SMG is a rare anomaly that can mimic other neck or salivary pathologies. High-resolution imaging is essential for diagnosis, while histopathology may be helpful when autoimmune disease is suspected. Awareness of this condition helps prevent unnecessary invasive procedures and supports appropriate conservative management.
Keywords. Ectopic Submandibular Gland, Acalculous Sialadenitis, Cervical Mass, MRI Neck.

Introduction

The submandibular glands (SMGs) are major salivary glands responsible for oral lubrication, antimicrobial defence, and mucosal protection via the saliva-secreting system. Each gland drains saliva via the Wharton duct into the floor of the mouth at the sublingual caruncle [1]. Anatomically, the SMGs are normally located in the submandibular triangle, beneath the jaw, and are divided by the mylohyoid muscle into superficial and deep lobes. During embryonic development, the SMGs arise from the oral epithelium, while their surrounding stroma is derived from neural crest cells. in glandular development or migration, it can lead to anomalies such as aplasia, heterotopia, or ectopia [1,2].

Ectopic SMG is an extremely rare developmental anomaly, with only a few cases reported in the literature. [1-3]. Patients with ectopic SMG may be asymptomatic or present with mimicking pathological neck masses, potentially leading to misdiagnosis or unnecessary interventions. Moreover, the SMGs are vulnerable to inflammatory, obstructive, and neoplastic conditions and may also be displaced during head and neck surgery. We report a rare case of a right-sided ectopic SMG, highlighting the clinical implications for diagnosing such cases.

Case presentation

A 52-year-old female presented to our outpatient clinic with a one-year history of chronic mouth dryness and intermittent dysphagia. She had no history of salivary gland surgery or intervention, and her past medical history was unremarkable. On physical examination, her vital signs were within normal limits. Neck examination revealed no palpable masses, tenderness, or asymmetry. Laboratory investigations showed normal complete blood count (CBC), thyroid function tests, and inflammatory markers (C-reactive protein and erythrocyte sedimentation rate). Initial evaluation with neck ultrasound (US) revealed displacement of the right SMG from its normal anatomical location, consistent with an ectopic SMG. Associated with bilateral reactive cervical lymphadenopathy. The parotid and thyroid glands, cervical vessels, and adjacent soft tissues appeared normal.

To further characterize the findings, a contrast-enhanced magnetic resonance imaging (MRI) scan of the neck was performed. The MRI confirmed a right-sided ectopic SMG, located laterally and superior to the thyroid gland (Figures 1&2). The right Wharton's duct was dilated, but no evidence of calculi or obstructive lesions was observed. The gland appeared hyperaemic and oedematous, consistent with acalculous sialadenitis. The gland was associated with bilateral reactive cervical lymphadenopathy. The oropharyngeal floor muscles were bilaterally symmetrical and normally developed, and no abnormalities were noted in the pharynx, larynx, parotid glands, or cervical vessels.

Given the combination of xerostomia, cervical lymphadenopathy, and the finding of the MRI suggestive of chronic sialadenitis, autoimmune disease such as Sjögren's syndrome was clinically suspected. Therefore, a minor salivary gland lip biopsy was performed. Histopathological examination revealed mild lymphocytic infiltration with preserved acinar and ductal architecture, effectively ruling out Sjögren's syndrome. As no obstructive or autoimmune pathology was identified, surgical intervention was not indicated. The patient

Received*: 26-09-2025 -* ***Accepted****: 24-11-2025 -* ***Published****: 03-12-2025*

was managed conservatively with symptomatic treatment and scheduled for regular follow-up to monitor glandular function and symptom progression.

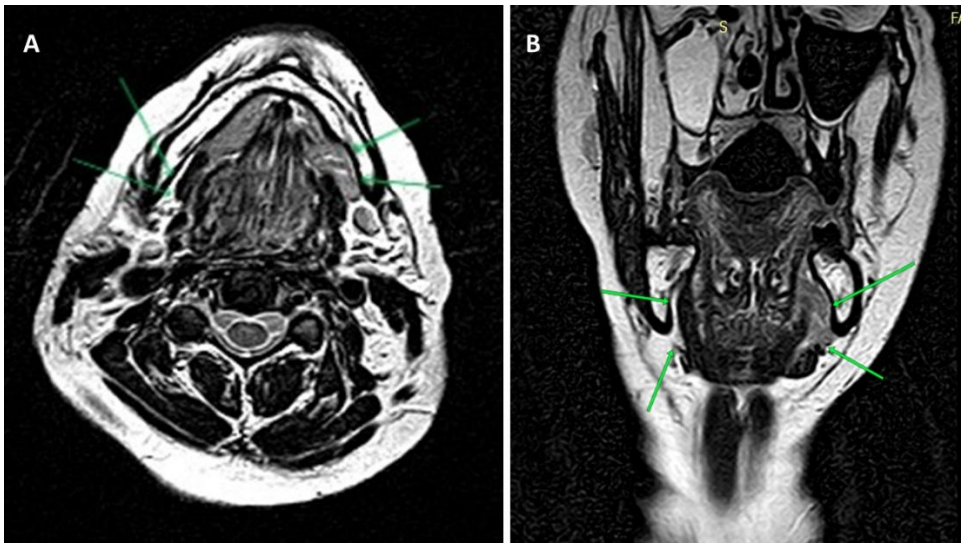

Figure 1. Axial and coronal T2-weighted MRI images demonstrate an empty submandibular fossa on the right side

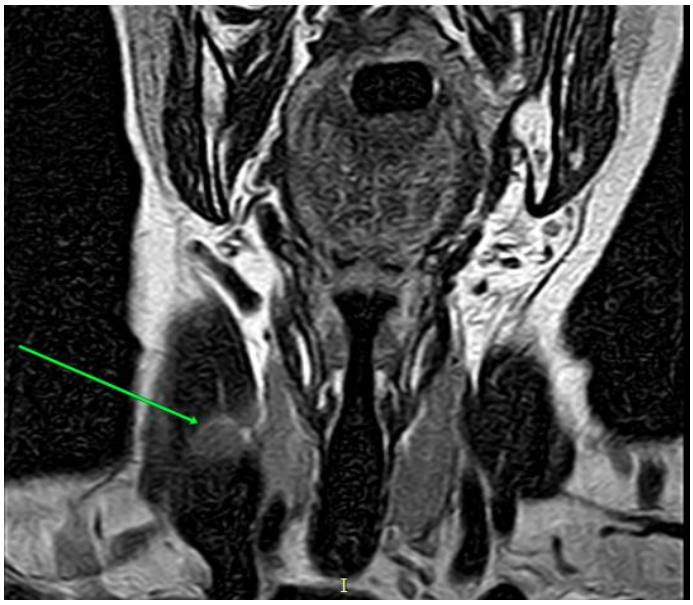

Figure 2. Coronal T2-weighted MRI shows the ectopic right submandibular gland positioned laterally and superior to the thyroid gland

Discussion

Ectopic SMGs are uncommon congenital anomalies resulting from the aberrant development or migration of glandular tissue during embryogenesis. Among the major salivary glands, ectopic SMGs are particularly rare, with only a limited number of cases reported in the literature. [1-3]. In the present case, the right SMG was located laterally and superior to the thyroid gland and associated with ductal dilatation and acalculous sialadenitis.

SMG develops during the sixth week of gestation from an epithelial proliferation in the floor of the mouth, which migrates posterolateral beneath the mylohyoid muscle [4]. Arrested migration, abnormal developmental pathways, or displacement are the main causes of ectopic positioning. Heterotopic salivary tissue has been reported in various head and neck locations, including the mandible, parapharyngeal space, middle ear, thyroid capsule, and along the anterior border of the sternocleidomastoid muscle [4-6]. SMG in the lower neck is particularly rare.

The clinical presentation of ectopic SMG varies depending on the size, location, and degree of ductal drainage impairment. Ectopic SMG can present clinically as asymptomatic or with symptoms like pain, swelling, dysphagia, or recurrent sialadenitis [7]. Chronic dry mouth and intermittent dysphagia impair salivary drainage and cause inflammatory changes. Imaging revealed dilatation of Wharton's duct without evidence of obstruction, suggesting an inflammatory rather than obstructive aetiology.

Received*: 26-09-2025 -* ***Accepted****: 24-11-2025 -* ***Published****: 03-12-2025*

Imaging plays a key role in the diagnosis of ectopic SMG. The US is a valuable initial modality for providing a noninvasive and readily available assessment of gland morphology and ductal anatomy. In this patient, US demonstrated displacement of the right SMG with preserved echotexture and no evidence of calculi, suggesting an ectopic location. MRI offers superior soft-tissue contrast and can accurately diagnose and confirm the ectopic location of SMG, and also assess ductal anatomy, and differentiation from other pathologies such as lymphadenopathy, ectopic thyroid tissue, branchial cleft anomalies, thyroglossal duct cysts, lipomas, or salivary gland neoplasms. In our patient, MRI confirmed the ectopic location and excluded obstructive or neoplastic disease.

Given the overlapping features between chronic sialadenitis and autoimmune disorders, histopathological evaluation remains essential for a definitive diagnosis. Histopathology may be useful when autoimmune disorders such as Sjögren's syndrome are suspected, as in our patient's minor salivary gland biopsy, which showed lymphocytic infiltration without features of autoimmune sialadenitis. The absence of other congenital anomalies helped exclude syndromic associations to major salivary gland aplasia, such as Treacher–Collins syndrome, ectodermal dysplasia, or lacrimo-auriculo-dento-digital (LADD) syndrome [8].

Management of ectopic SMG cases depends on symptom severity, underlying pathology, and the risk of complications [9]. Asymptomatic patients may be managed with observation, while symptomatic cases may require medical treatment for inflammation or surgical excision in the presence of obstruction, neoplasia, or recurrent infection. In this case, the absence of calculi, neoplastic changes, or autoimmune disease supported a conservative approach with symptom-based therapy and routine follow-up.

Conclusion

Ectopic SMG is a rare developmental anomaly that can resemble other cervical or salivary pathologies. Accurate diagnosis relies on careful clinical evaluation and high-resolution imaging, with histology used only when necessary. This case underscores the importance of considering ectopic SMG in the differential diagnosis of unusual neck masses to avoid unnecessary invasive procedures and ensure proper management.

Conflict of interest

There are no competing interests of any of the authors, and they have no connection to the industry or organizations.

Funding sources

There was no specific grant for this research from any funding organization, the private sector, or the not-for-profit sector.

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Received*: 26-09-2025 -* ***Accepted****: 24-11-2025 -* ***Published****: 03-12-2025*