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Case Report

A case report of ectopic salivary gland tissue in the tongue base ☆,☆☆

*Jin Kai Soh, MBChB**, *Bushra Awan, FRCR, MBBS*

Department of Radiology, Salisbury District Hospital, Salisbury NHS Foundation Trust, Odstock Road, Salisbury SP2 8BJ, United Kingdom

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ABSTRACT

Ectopic salivary gland tissue is the presence of normal salivary gland tissue developing outside of the major and minor salivary glands. We present a rare, incidental case of lingual ectopic salivary gland tissue in a 52-year-old male with known Birt-Hogg-Dubé syndrome, diagnosed on imaging as tongue mass. Excisional biopsy of the left tongue base lesion was performed and histologic examination revealed ectopic salivary gland tissue. We found only a few cases in literature of ectopic salivary gland tissue situated in the tongue. This case illustrates the need to consider ectopic salivary gland tissue as part of the differential diagnoses of a tongue mass.

Introduction

Heterotopic salivary gland tissue (HSGT), also known as "ectopic salivary gland," or "salivary gland choristoma," is defined as the presence of normal salivary gland tissue occurring outside the normal distribution of the major (parotid, submandibular, and sublingual) and minor salivary glands of the oral cavity, pharynx, and upper airways [1]. The salivary gland acini cells are present in any abnormal location without any duct system. In this case report, we describe a case of ectopic salivary gland tissue within a rare location of the tongue from a patient with a background of Birt-Hogg-Dubé syndrome. This incidental finding was first noticed on imaging as a tongue mass and excisional biopsy was performed for histologic analysis.

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* Corresponding author.
E-mail address: alistairsoh@gmail.com (J.K. Soh).

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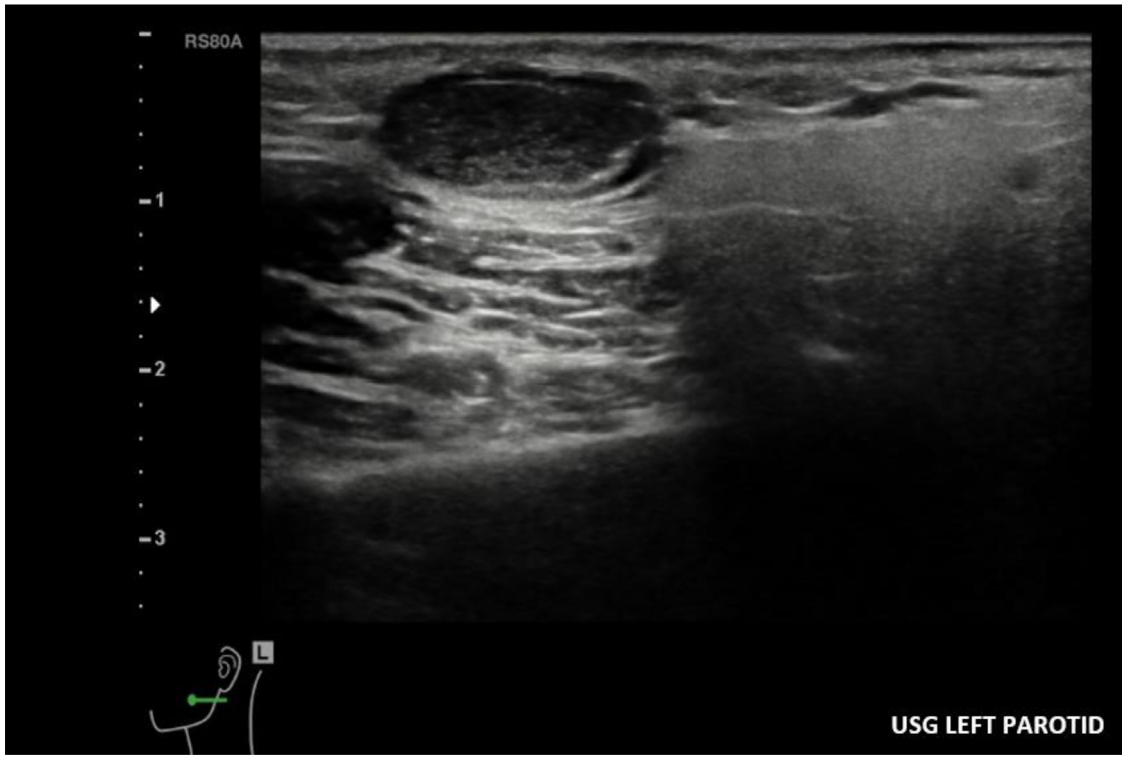

Fig. 1 – Ultrasound guidance (USG): long axial view of a well-defined subcutaneous lesion in the left parotid region, appearing hypoechoic.

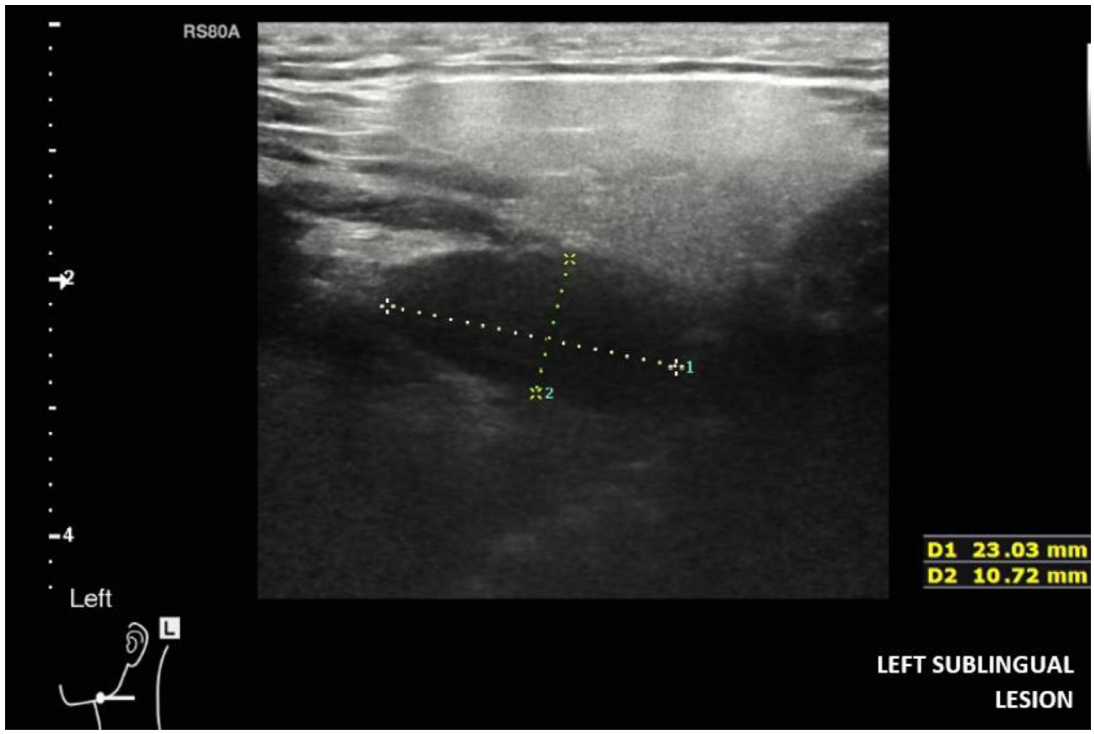

Fig. 2 – Ultrasound: transverse view of the left submandibular region showing a well-defined hypoechoic area deep to the submandibular gland, suggestive of tongue lesion.

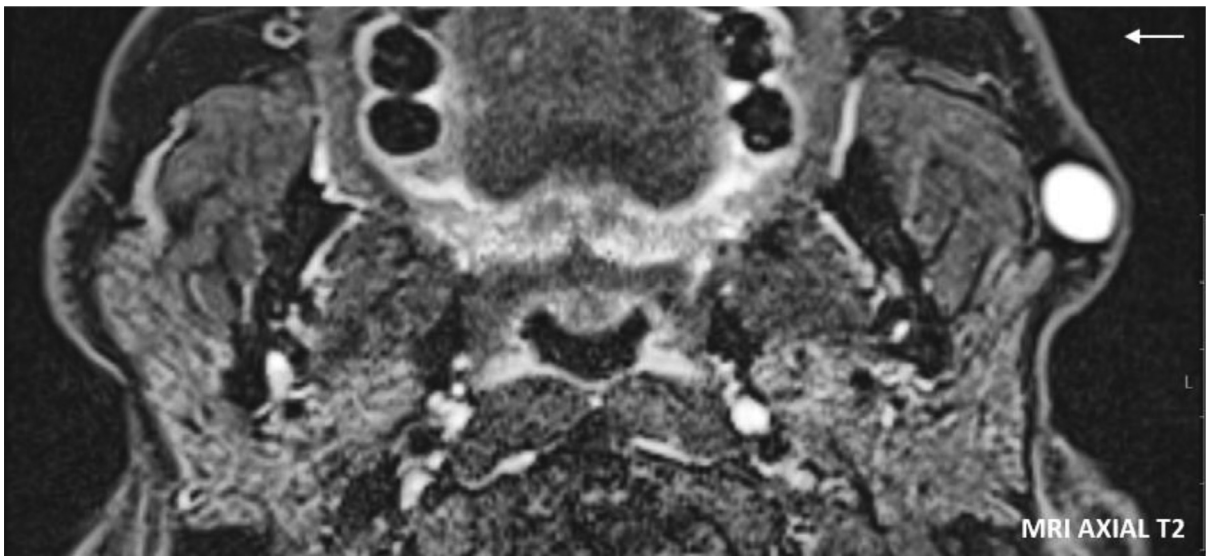

Fig. 3 – MRI: Axial T2 fat-saturated sequence. A well-defined T2 hyperintense subcutaneous lesion in the left parotid region.

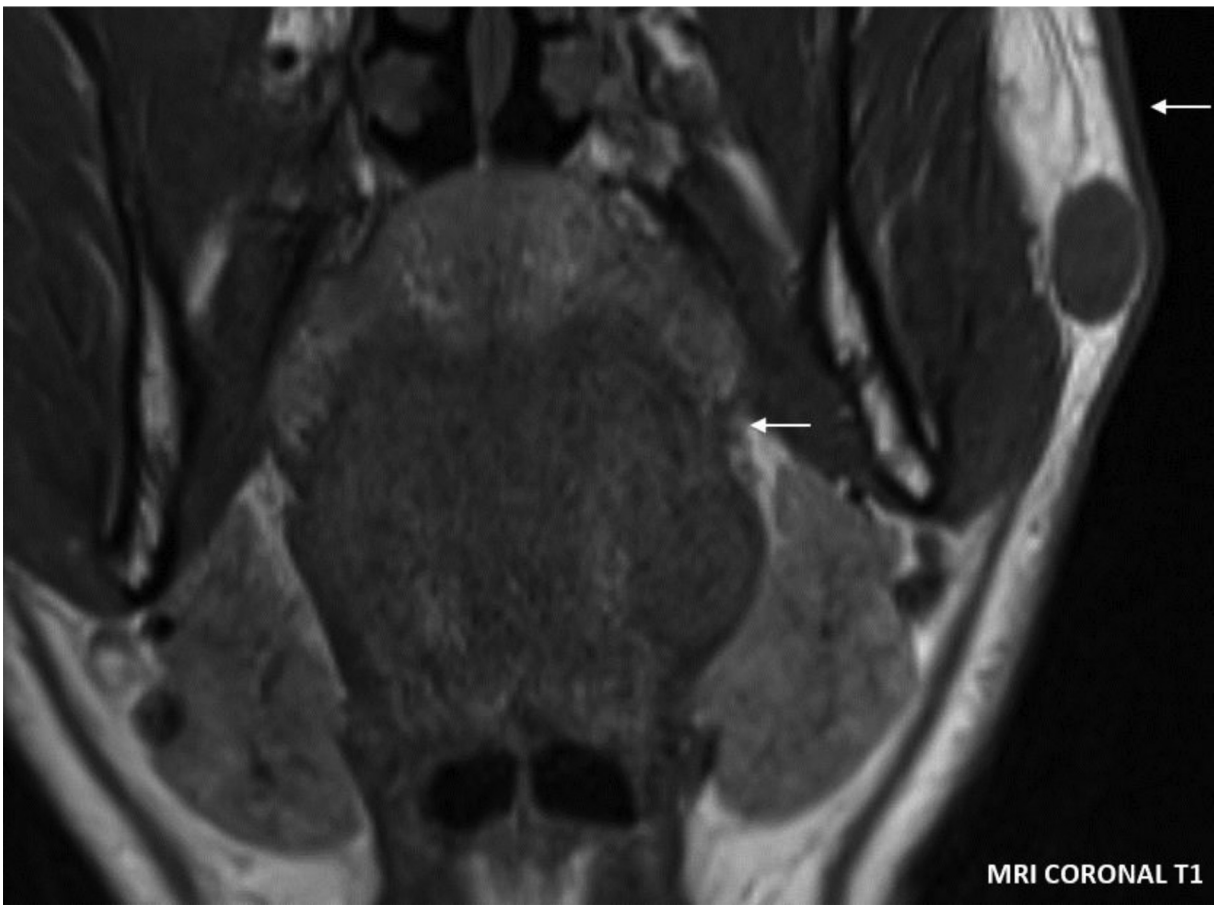

Fig. 4 – MRI: Coronal T1 sequence. A well-defined T1 hypointense subcutaneous lesion in the left parotid region, and another well-defined T1 hypointense ovoid lesion in the left tongue.

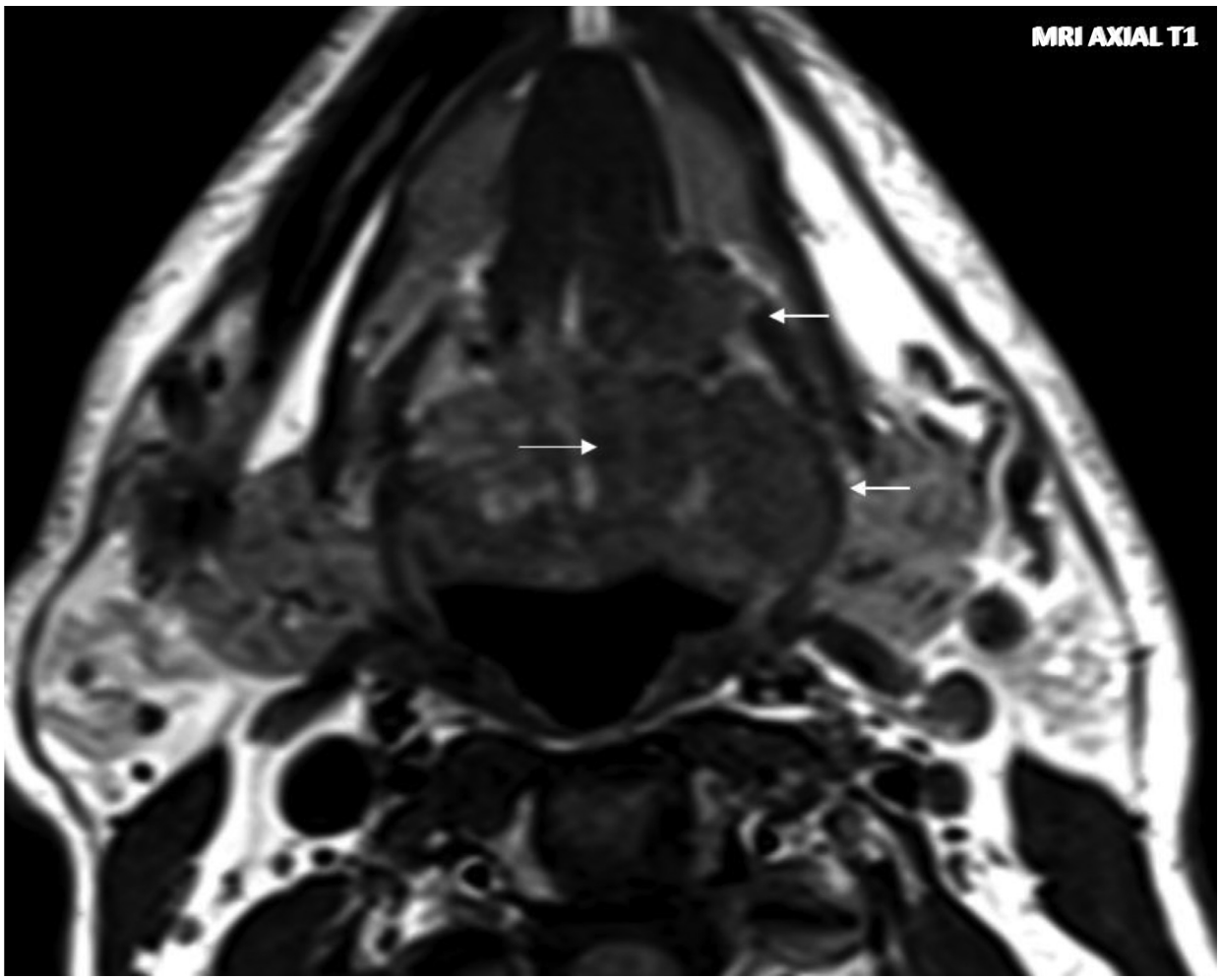

Fig. 5 – MRI: Axial T1 sequence. Three well-defined T1 hypointense lesions in the left tongue.

Table 1 – Review in literature of other cases of ectopic salivary gland tissue in the tongue.

Author	Year	Country	Localization	Side	Age	Sex	Treatment
Guerrissi [14]	2000	Argentina	Hyoid region	M	26 years	F	Surgical excision (Sistrunk operation)
Mehmet et al. [15]	2014	Turkey	Tongue base	N/A	Newborn (40-week gestation)	M	Surgical resection (bipolar electrocautery)
Meng et al. [16]	2014	China	Tongue base	N/A	1 year	M	Surgical resection (en bloc resection)
Hiebert et al. [17]	2016	USA	Tongue base	M	Newborn (41-week gestation)	F	Surgical resection
Trieu et al. [18]	2020	USA	Lateral tongue base	L	6 months	M	Transcervical excision

Case report

A 52-year-old gentleman was presented to our hospital with a 2-year history of intermittent nonspecific pain involving angle of the left mandible extending up toward the vertex, occiput, and out towards the ipsilateral shoulder. He also noted difficulty with head rotation, ipsilateral eye and ear pain. Physical examination was notable for a well-defined superficial lump on the left side of the face, lying immediately below the skin anterior to the parotid gland.

This patient has a past medical history of recently diagnosed Birt-Hogg-Dubé syndrome (BHDS), irritable bowel syndrome, and depression. He has a known family history of BHDS, with both his brother and sister tested positive for BHDS which he suspects to be inherited from his mater-

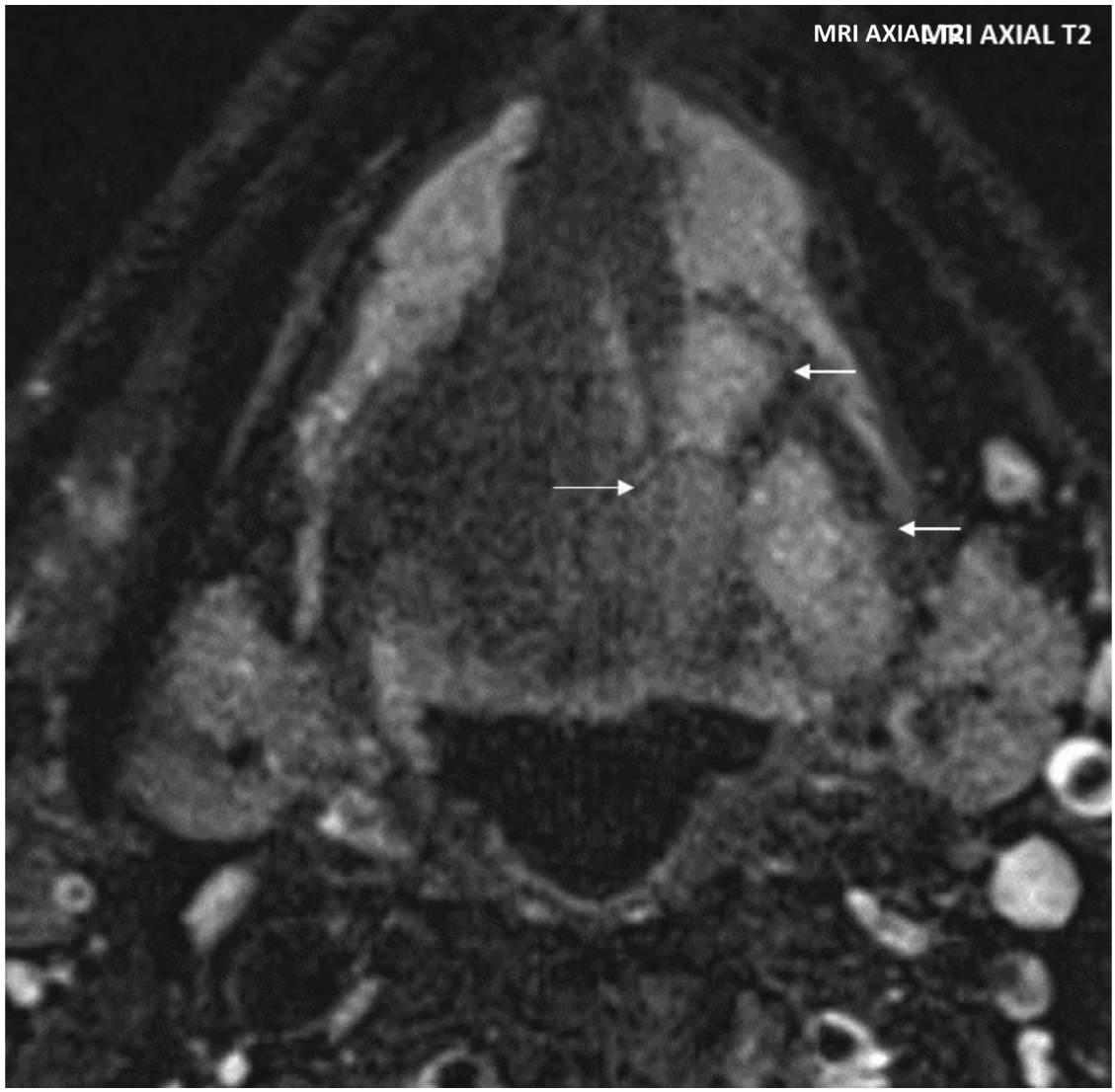

Fig. 6 – MRI: Axial T2 fat-saturated sequence. Three well-defined T2 hyperintense lesions in the left tongue.

nal side. Previous high-resolution computed tomography (CT) chest confirmed the presence of multiple thin-walled cystic areas predominantly in the lower lobes bilaterally in a paraseptal distribution, atypical for smoking-related emphysema. He was later tested positive for the pathogenic variant on FLCN gene analysis. He receives an annual MRI of the urinary tract for surveillance of renal malignancy. The patient has also received cryotherapy and ablative laser therapy for his multiple facial cutaneous fibrofolliculomas.

Patient was referred for ultrasound for the left face subcutaneous palpable lump. Ultrasound showed a well-defined solid tissue lump. Ultrasound-guided fine needle aspiration (FNA) results of the subcutaneous small lesion were inconclusive. The initial impression was a pleomorphic adenoma of the left parotid gland (Fig. 1).

Additionally, a more defined left sublingual region lesion was incidentally identified on the ultrasound. It was a difficult-to-examine region, so no definitive comment was made on the report and clinical examination of the tongue base and sublingual region was suggested (Fig. 2).

A magnetic resonance imaging (MRI) of the parotid gland was then performed which confirmed a solitary, well-defined superficial lesion anterior to the margin of the left parotid gland subcutaneously, measuring at 18 × 12 × 10 mm (Fig. 3).

There were also 3 separate enhancing lesions identified along the left half of the tongue, the largest of which is a 23 × 12 mm well-defined T1 hypointense and T2 hyperintense region lying along the inferolateral aspect of the left half of the tongue deep to the hyoglossus. All 3 lesions had no obvious infiltration of the extrinsic muscle of the tongue (Figs. 4-7).

A positron emission tomography–computed tomography (PET-CT) of the lesion was performed following the MRI, as a sinister tongue lesion such as SCC could not be excluded.

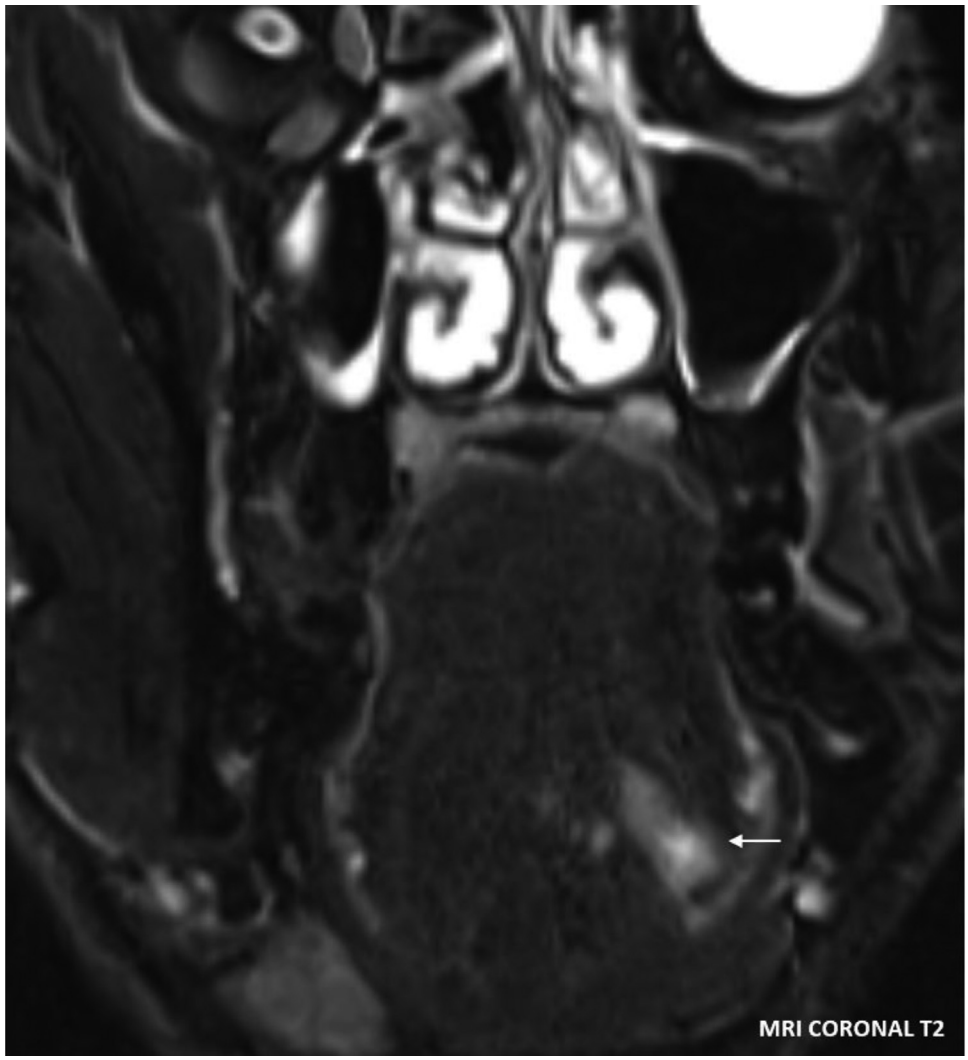

Fig. 7 – MRI: Coronal T2 fat-saturated sequence. Well-defined, ovoid T2 hyperintense lesion in the left tongue.

The well-defined soft tissue anterior to the left parotid gland was not overtly avid. However, high activity was noted within the left glossotonsilar sulcus extending to the inferolateral aspect of the left tongue, metabolic measurement of 23 mm/SUV max 9.3. Hypermetabolic foci also present anteromedially in the other 2 lesions (Figs. 8 and 9).

The patient then underwent an excisional biopsy of the left tongue base mass under general anesthesia. Histologic analysis of the tissue specimens taken from the left tongue base mass and deep tissue within left tongue base demonstrated minor salivary gland elements as well as salivary tissue embedded within the muscle, with no evidence of neoplasia. The findings were consistent with a diagnosis of ectopic salivary gland tissue that was, however, not contributing to the patient's core symptoms. A further MRI of the cervical spine confirmed the presence of osteophytes narrowing of the left vertebral foramen at spinal levels C2-C3, C3-C4, C4-C5, and C5-C6 giving rise to his radicular pain.

Discussion

HSGT has been well documented in numerous locations throughout the head and neck have been documented in literature such as the mandible, middle ear, palatine tonsil, gingiva, pituitary gland, cerebellopontine angle, thyroid gland, and the larynx [2–9]. However, the most common site of predilection is the intraparathyroid lymph nodes [10]. There have been case reports of HSGT identified away from the head and neck such as the esophagus, rectum, and vulva [11–13]. Reported lingual involvement of HSGT is scarce. Our literature search revealed only 5 case reports that were either originally written in English, or translated to English, to date (Table 1). To the best of our knowledge, we report the sixth of its kind in literature.

Embryologic development of salivary gland tissue begins during the sixth week of gestation [19]. It is still unclear how

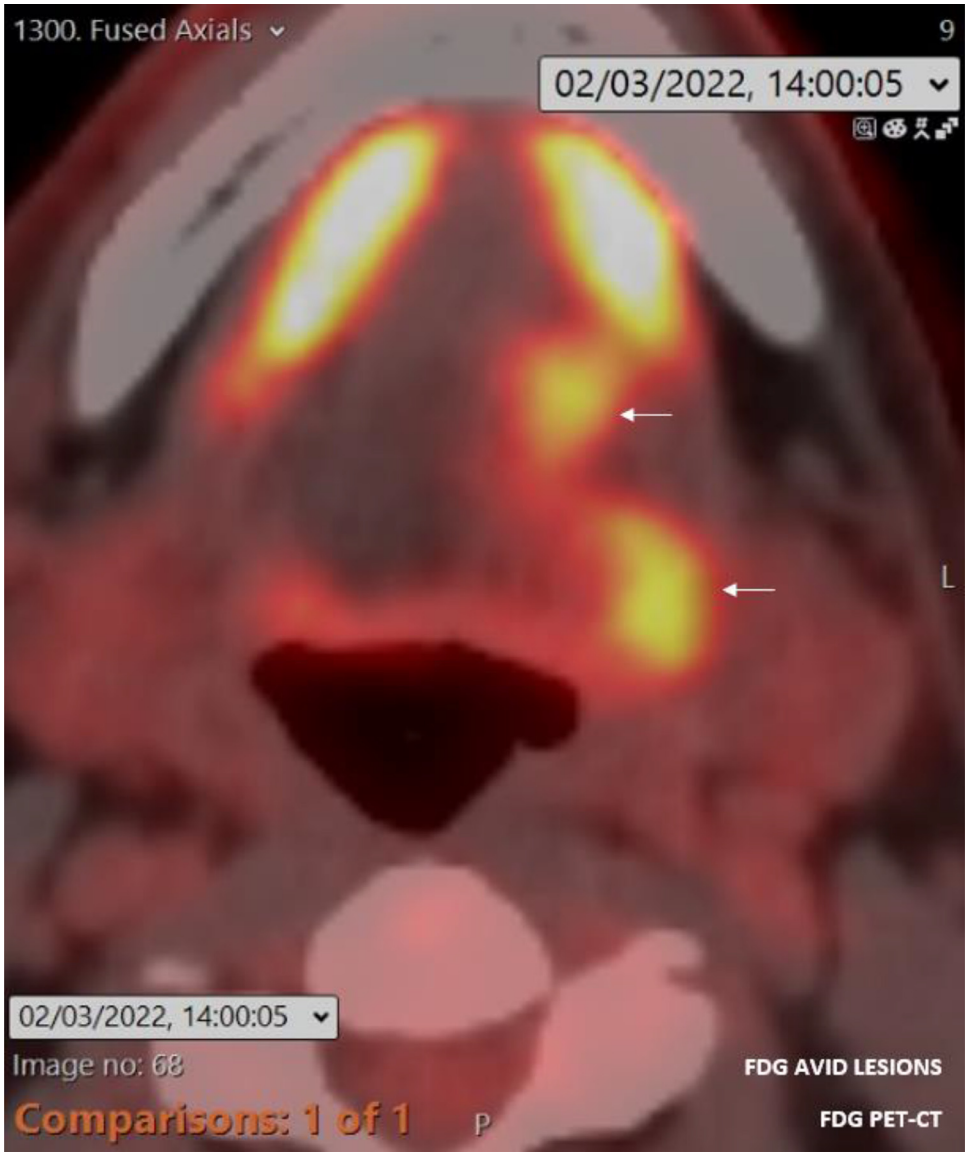

Fig. 8 – PET-CT: Axial fluorodeoxyglucose (FDG) PET-CT. Two significantly PET avid lesions seen in the left tongue.

salivary gland tissue that originated from oral epithelial cells migrate aberrantly to the base of the tongue.

The occurrence of salivary tissue neoplasm within ectopic salivary gland tissue is rare, with 80% of HSGT tumors being benign [20]. However, neoplasm has been reported in intraparotid and cervical lymph nodes [21]. Most tongue base tumors arise from the mucosa and are squamous cell carcinoma. Differential diagnosis of tongue base mass to consider are normal lymphatic tissue, ectopic thyroid tissue, lipoma, lymphoma, hemangioma, granular cell tumors, benign and malignant tumors of minor salivary gland origin, rhabdomyosarcoma, and metastatic disease [22]. Patients with lingual HSGT reported in literature were symptomatic, particularly among neonates that presented with respiratory symptoms. In our case, the patient's symptoms were caused by radiculopathy and HSGT was found incidentally.

As HSGT is essentially normal tissue, it generally does not require treatment if not otherwise symptomatic. Surgical excision can be performed for either histopathologic diagnosis, cosmetic purposes, or symptomatic treatment [23]. There have been no cases of recurrence or continuous growth reported even when the mass is incompletely removed [8].

This patient was recently diagnosed with Birt-Hogg-Dubé (BHD) syndrome, an autosomal dominant condition characterized clinically by multiple cutaneous fibrofolliculomas, pulmonary cysts, spontaneous pneumothoraces, and an increased risk of renal neoplasia, and less frequently salivary gland tumors [24,25].

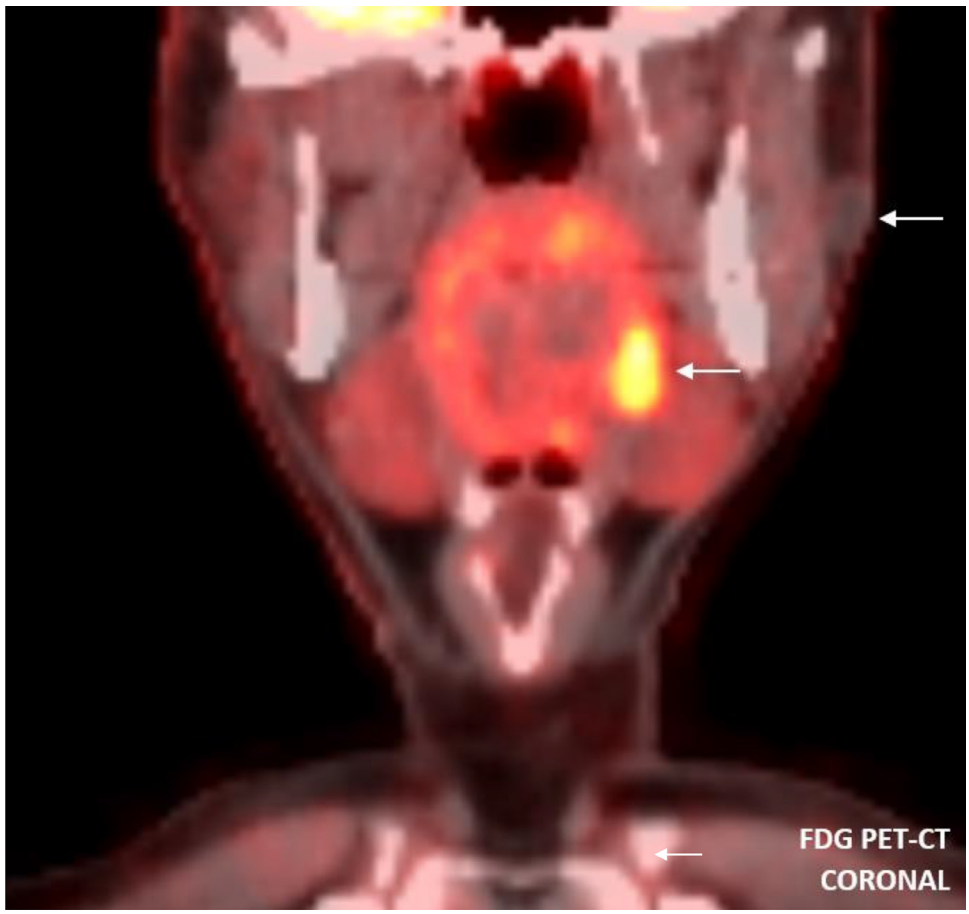

Fig. 9 – PET-CT: Coronal FDG PET-CT. A nonavid, well-defined left parotid lesion seen (arrow above). A significantly avid left tongue lesion seen (arrow below).

There is no correlation between BHD and ectopic tissue in current literature. However, several cases of salivary gland tumor development in BHD have been reported [26,27]. It is important to consider these presentations as differentials for a soft tissue facial mass in the context of BHD.

Patient consent

Written informed consent was obtained from the patient for publication of this case report, including accompanying images.

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