

Isolated aplasia of submandibular salivary gland and contralateral prominence of submandibular and sublingual salivary glands incidentally found on ⁶⁸Ga-prostate-specific membrane antigen positron emission tomography–computed tomography

ABSTRACT

Prostate-specific membrane antigen (PSMA)-targeted imaging is now an effective tool for the evaluation of prostate cancer patients. Although salivary glands take up ⁶⁸Ga-PSMA avidly, pathologies of these glands may be readily noticeable. Herein, we present a case of prostate cancer referred for ⁶⁸Ga-PSMA positron emission tomography–computed tomography in whom an isolated aplasia of the submandibular salivary gland was incidentally found.

Keywords: ⁶⁸Ga-prostate-specific membrane antigen positron emission tomography–computed tomography, isolated aplasia, submandibular salivary gland

INTRODUCTION

We present a case of prostate cancer referred for ⁶⁸Ga-prostate-specific membrane antigen (PSMA) positron emission tomography–computed tomography (PET-CT) in whom an isolated aplasia of the submandibular salivary gland was incidentally found.

CASE SUMMARY

A 68-year-old patient with prior adenocarcinoma of the prostate and then radical prostatectomy was referred for metastatic evaluation following a rise in the serum prostate-specific antigen level. After obtaining informed consent, the patient underwent a ⁶⁸Ga-PSMA PET-CT scan 60 min after intravenous administration of 208 MBq of ⁶⁸Ga-PSMA from the vertex to the upper thigh [Figure 1a], on which, in addition to residual or recurrent disease in the surgical bed of prostatectomy, the submandibular salivary gland was absent on the right side, and in contrast, the

contralateral one seemed prominent. The sublingual glands were also asymmetric [Figure 1b-d]. PSMA has become a

ABBAS YOUSEFI-KOMA, REYHANE AHMADI¹, SABA KARAMI GORZI², YASER SHIRAVAND¹, MOHSEN QUTBI¹

Chronic Respiratory Diseases Research Center, National Research Institute of Tuberculosis and Lung Diseases, Shahid Beheshti University of Medical Sciences, ¹Department of Nuclear Medicine, School of Medicine, Taleghani Educational Hospital, Shahid Beheshti University of Medical Sciences, ²Department of Medical Radiation Engineering, Science and Research Branch, Islamic Azad University, Tehran, Iran


Address for correspondence: Dr. Mohsen Qutbi, Department of Nuclear Medicine, Taleghani Hospital, Yaman St., Velenjak, Tehran 1985711151, Iran.
E-mail: mohsen.qutbi@gmail.com, mohsen.qutbi@sbm.ac.ir

Submission: 10-Feb-20, **Revised:** 01-May-20, **Accepted:** 21-May-20, **Published:** 22-Jul-20

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Yousefi-Koma A, Ahmadi R, Gorzi SK, Shiravand Y, Qutbi M. Isolated aplasia of submandibular salivary gland and contralateral prominence of submandibular and sublingual salivary glands incidentally found on ⁶⁸Ga-prostate-specific membrane antigen positron emission tomography–computed tomography. *World J Nucl Med* 2020;19:322-3.

Access this article online	
Website: www.wjnm.org	Quick Response Code 
DOI: 10.4103/wjnm.WJNM_13_20	

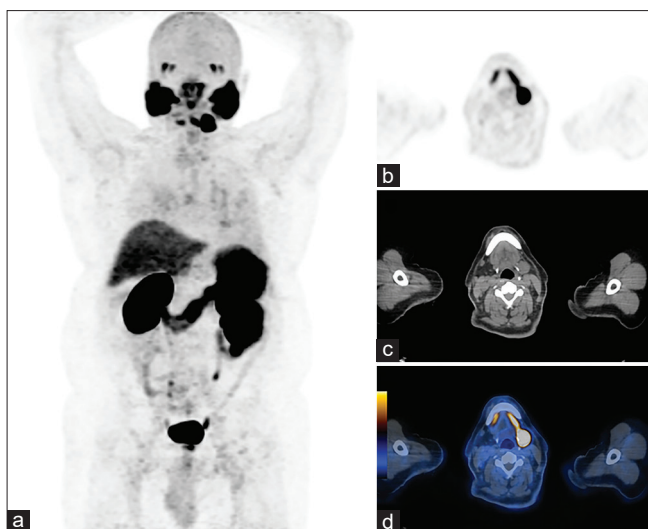


Figure 1: Anterior maximum intensity projection image (a) of ^{68}Ga -prostate-specific membrane antigen positron emission tomography-computed tomography shows absent submandibular salivary gland on the right side as well as prominence of the contralateral one. The absent gland is better depicted on ^{68}Ga -prostate-specific membrane antigen positron emission tomography (b), computed tomography (c), and fused positron emission tomography-computed tomography image (d)

favorable molecular target for imaging of the prostate cancer tissues, and to date, PSMA-based PET scanning has been successfully incorporated into the clinical practice. Despite its distinct advantages in the diagnosis and therapy of malignant prostate tissues, the use of such target for imaging has been challenged, to some extent, as a result of a diverse range of false-positive and incidental prostate-unrelated findings.^[1-3] Salivary glands normally demonstrate intense uptake on ^{68}Ga -PSMA PET-CT. Either decrease or absence of uptake, particularly when unilateral, may be a clinically significant finding and deserves attention. Congenital absence of salivary glands is rare and mostly multiple glands are involved. Unilateral “isolated” aplasia of submandibular salivary gland is an exceptionally rare finding. It may be associated with other anomalies such as hemifacial microsomia and mandibulofacial dysostosis.^[4-7] The biologic mechanism is not clearly known, but may result from defect in fibroblast growth factor signaling pathways that prevents development of salivary gland tissue during embryogenesis.^[8,9] On the other hand, the contralateral submandibular salivary gland, as in our patient, or sometimes ipsilateral or contralateral sublingual salivary glands may show more prominence, possibly as a compensatory hypertrophy against xerostomia.^[5,6] This issue, in turn, poses interpretive challenges because of pathology of the gland itself or obscuring adjacent (e.g., mandibular) PSMA-avid lesions.^[10]

CONCLUSION

^{68}Ga -PSMA PET-CT imaging as a promising modality for

the evaluation of patients with prostate cancer is being incorporated into clinical practice. Familiarity with pitfalls, limitations, and also varieties of incidental findings may be of importance for clinicians.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Sheikhabaei S, Werner RA, Solnes LB, Pienta KJ, Pomper MG, Gorin MA, *et al.* Prostate-specific membrane antigen (PSMA)-targeted PET imaging of prostate cancer: An update on important pitfalls. *Semin Nucl Med* 2019;49:255-70.
2. Farag M, Bolton D, Lawrentschuk N. Prostate-specific membrane antigen for the surgical oncologist: Interpreting expression beyond the prostate. *ANZ J Surg* 2020;90:715-8.
3. Van de Wiele C, Sathekge M, de Spiegeleer B, de Jonghe PJ, Beels L, Maes A. PSMA-targeting positron emission agents for imaging solid tumors other than non-prostate carcinoma: A systematic review. *Int J Mol Sci* 2019;20:4886.
4. Yan Z, Ding N, Liu X, Hua H. Congenital agenesis of all major salivary glands and absence of unilateral lacrimal puncta: A case report and review of the literature. *Acta Otolaryngol* 2012;132:671-5.
5. Srinivasan A, Moyer JS, Mukherji SK. Unilateral submandibular gland aplasia associated with ipsilateral sublingual gland hypertrophy. *AJNR Am J Neuroradiol* 2006;27:2214-6.
6. Kara M, Güçlü O, Dereköy FS, Resorlu M, Adam G. Agenesis of submandibular glands: A report of two cases with review of literature. *Case Rep Otolaryngol* 2014;2014:569026.
7. Ahmed M, Strauss M, Kassaie A, Shotelersuk V, DeGuzman R. Bilateral submandibular gland aplasia with clinico-radiological mass due to prolapsing sublingual salivary tissue through mylohyoid boutonniere: A case report and review. *Dentomaxillofac Radiol* 2009;38:121-4.
8. Jaskoll T, Abichaker G, Witcher D, Sala FG, Bellusci S, Hajhosseini MK, *et al.* FGF10/FGFR2b signaling plays essential roles during in vivo embryonic submandibular salivary gland morphogenesis. *BMC Dev Biol* 2005;5:11.
9. Jaskoll T, Witcher D, Toreno L, Bringas P, Moon AM, Melnick M. FGF8 dose-dependent regulation of embryonic submandibular salivary gland morphogenesis. *Dev Biol* 2004;268:457-69.
10. Lengana T, Lawal I, Boshomane T, Ololade K, Reyneke F, Kaoma C, *et al.* Salivary gland activity obscures mandibular metastasis of prostate carcinoma on ^{68}Ga -prostate-specific membrane antigen PET. *Clin Nucl Med* 2018;43:106-9.