

CASE REPORT

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Oral melanotic hyperpigmentation following external beam radiotherapy for mucoepidermoid carcinoma of the parotid gland

Joshua B Kaelberer, Keaton A Rummel, John M Watkins

ABSTRACT

Introduction: Epithelial hyperpigmentation from is a commonly encountered side effect from radiation therapy applications, most frequently described in skin. We here report a case of radiotherapy-associated oral hyperpigmentation.

Case Report: A 46-year-old female with parotid mucoepidermoid carcinoma underwent resection with involved margins, followed by adjuvant radiotherapy. During her course of treatment, non-tender, non-confluent, well-demarcated palatal and lingual hyperpigmented macules were noted, monitored without intervention. Serial images demonstrate gradual resolution during several months post-treatment.

Conclusion: Oral hyperpigmentation is likely an underreported phenomenon, and appears to be largely cosmetic, with stigmata resolving gradually over time.

Keywords: Hyperpigmentation, Oral cavity, Radiotherapy

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INTRODUCTION

Mucoepidermoid carcinomas are tumors that arise from mucous-secreting and squamous cells within salivary glands, most commonly the parotid gland [1]. Demonstrating a slight female preponderance and most commonly presenting in middle age, most tumors are low- to intermediate-grade, with a small proportion demonstrating high-grade features. Wide local excision or parotidectomy, with or without neck dissection, is recommended as primary intervention, with adjuvant radiotherapy considered in the context of involved surgical margin(s), perineural invasion, intraoperative tumor spill, high-grade histology, and/or lymph node involvement [2].

CASE REPORT

During a routine check-up, a healthy 46-year-old Pacific Islander woman was found to have a new, painless mass on her right parotid gland, which she had not noticed earlier. She did not feel any fullness, swelling, or facial weakness. Subsequent computed tomography (CT) imaging characterized the lesion as a 2.6 cm complex solid/cystic mass with mild internal and rim enhancement and suggestion of subdermal extension. Biopsy demonstrated well- to moderately differentiated mucoepidermoid carcinoma, with minimal atypia and predominance of mucous cells. Following head/neck surgeon consultation, total parotidectomy plus lymph node dissection was recommended, as is routine for this diagnosis [1]. Pathology demonstrated a 3.3 cm primary tumor with microscopic extraparenchymal invasion, involvement of superficial and deep surgical

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margins, without involvement of 29 dissected nodes (pT2N0 R1). Due to surgical margin involvement, an established risk factor for local recurrence [1, 2], which can be associated with additional morbidity from re-resection. As such, the patient was referred to radiation oncology for consideration of adjuvant radiotherapy, which has demonstrated efficacy in reducing the risk of local recurrence [3]. Standard fractionation was elected, prescribed as 66 Gy in 33 once-daily fractions to the unilateral resection bed plus margin, as described elsewhere [4]. A 6 MV photon volumetric modulated arc technique was employed in order to optimize target volume coverage while minimizing unnecessary dose to adjacent organs at-risk (and associated acute and late toxicity risks), as is standard for head/neck radiotherapy [5]. Specifically, the mean oral cavity dose was 11.3 Gy and the maximum oral cavity dose was 22.8 Gy, with mandible, spinal canal, pharyngeal constrictors, and contralateral parotid gland meeting treatment plan objectives. Isodose map is demonstrated in Figure 1.

Just over two weeks into radiotherapy, the patient inquired about new well-demarcated patches of painless oral darkening, noted primarily on the tongue, palate, and right buccal mucosa (Figure 2A). These remained unchanged during the remainder of her treatments, painless and without oral discomfort or mucositis, and focal oral melanocytic hyperpigmentation was suspected (Figure 2B). Excepting anticipated moderate skin xeroderma with dry desquamation, moderate xerostomia, and dysgeusia, the patient completed her course of treatment and remained asymptomatic from these macules, which gradually faded over time, without intervention or complication (Figure 2C and D). Skin changes, oral dryness, and taste sensation gradually resolved over 3–6 months as well, and she remains without evidence of recurrence presently.

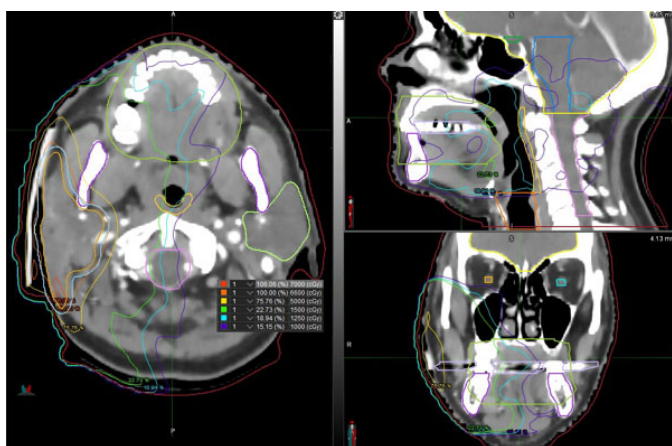


Figure 1: Radiotherapy isodose map in representative axial, sagittal, and coronal planes.



Figure 2: Patient's clinical exam demonstrating hyperpigmentation at (2.5 weeks into prescribed course of radiotherapy (A), end of treatment (B), four months post-treatment (C), and eight months post-treatment (D).

DISCUSSION

Melanotic hyperpigmentation of the tongue and oral mucosa secondary to radiotherapy appears to be an uncommon/underreported phenomenon, as there are few reports of similar cases within the medical literature [6, 7]. These cases demonstrated similar features, with otherwise asymptomatic macular hyperpigmentation, not associated with mucosal denudement, discomfort, or long-term pigmentation changes.

CONCLUSION

Oral hyperpigmentation is likely an underreported phenomenon, and appears to be largely cosmetic, with stigmata resolving gradually over time.

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Author Contributions

Joshua B Kaelberer – Conception of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Keaton A Rummel – Conception of the work, Design of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

John M Watkins – Conception of the work, Design of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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