Black Pigmentation of Tongue Following Radioactive Iodine Therapy: A rare Case Report

[1] Md. Sabbir Hossen, [2] Nafisa Jahan, [3] Ferdous Sharmin , [3]Nusrat Jahan

[1]1Medical Officer, Institute of Nuclear Medicine and Allied Sciences, Barishal, Bangladesh.
[2]Chief Medical Officer & Director, Institute of Nuclear Medicine and Allied Sciences, Barishal, Bangladesh
[3] Senior Medical Officer, Institute of Nuclear Medicine and Allied Sciences, Barishal, Bangladesh

Correspondence Address : Dr. Md. Sabbir Hossen Medical Officer, Institute of Nuclear Medicine & Allied Sciences, Sher-E-Bangla Medical College Campus, Barishal , Bangladesh. Email: sabbir714456@gmail.com

ABSTRACT

Radioactive iodine therapy (RAIT) is an effective method of treatment for differentiated thyroid carcinomas after a total or near-total thyroidectomy. Common side effects of this therapy are nausea, sialadenitis, dry mouth, etc.

Here we present an extremely rare case of black pigmentation of the tongue in a 28-year-old woman following RAIT after a total thyroidectomy due to papillary thyroid carcinoma (PTC), who presented with black discoloration of the dorsal surface of the tongue following the 3rd day of RAIT. Though the cause of discoloration remains unknown and there was a gradual reduction of dark color, a report should be done about this self-limiting, unusual side effect, which appeared to be apparently non-harmful.

Keywords: Black Pigmentation, Radioactive Iodine (RAI), Side effects.

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INTRODUCTION

Radioactive Iodine therapy (RAIT) is an important and principle efficacious form of treatment for differentiated thyroid carcinomas (DTC) following total or near total thyroidectomy. Most common early side effects of this therapy are nausea, sialadenitis, dry mouth and changes in the taste sensation, salivary gland pain, salivary gland swelling and vomiting (1-4). Black pigmentation of tongue following RAI therapy is an extremely rare side effect and there are very few cases documented worldwide till now, we found only two reported cases (5,6). Exact etiology of back pigmentation of tongue following RAI therapy is unknown. This is the first documented case of tongue pigmentation change following RAIT among the institutes treating DTC in Bangladesh, so, we took the opportunity of a novel documentation.

CASE REPORT

A 28-years old apparently healthy, married, woman, mother of a single child was referred to Institute of Nuclear Medicine & Allied Sciences (INMAS), Barishal on 14th post operative day for RAIT following total thyroidectomy with neck lymph node dissection due to PTC with cervical lymph node metastasis. Her clinical staging was pT2N1M0 and pre-therapy investigation revealed: Raised thyroid stimulating hormone (TSH)-23.48 µIU/ml (ref :0.3-5.0 µIU/ml), normal Thyroglobulin (Tg)-0.09 ng/ml (ref : <50 ng/ml), serum anti-TG antibody-51.57 IU/ml (ref : <100 IU/ml), serum calcium-9.90 mg/dl, random blood sugar (RBS)-5.06 mmol/L, serum creatinine-0.79 mg/dL. High resolution neck ultrasound (HRUS) showed no visible residual thyroid tissue in thyroid bed. Other biochemical studies like serum calcium, parathormone and vitamin D were unremarkable.

Patient was administered an ablation dose of 100 mci ^{131}I orally (TNM classification: pT2N1Mx with Stage-I according to AJCC 8th) after proper counseling of radiation safety issues. On the third day, she reported painless black discoloration of the dorsal surface of the tongue, loss of all special sensations except spicy, and numbness of the right upper limb from the shoulder and also in the right lower limb.

On a detailed history, she denied changes in the general sensation of the tongue, dry mouth, halitosis, sialorrhoea, pain, or swelling of the salivary glands. There was no history of changes in her oral hygiene regimen or diet; there was no history of taking colored lozenges, food or drinks. Previous

medical history was uneventful as well. However, levothyroxine, calcium with vitamin D and vitamin C were the ongoing medications at the time of developing this symptom. There was no history of taking oral contraceptives, alcohol or tobacco with no known allergy to any drug or contrast dye.

After the post-therapy whole body scan (RxWBS), a thorough physical examination was done, and an apparently healthy young female with no significant general or systemic changes was observed. Neurological examinations, especially of the upper and lower limbs, revealed no abnormalities. Oral cavity examination revealed blackening of the dorsal surface of the tongue along with prominent filliform papillae (Figure 1A), but no pigmentation was present in the ventral surface of the tongue, gums, or buccal mucosa. Oral hygiene, including teeth, gums, and palate, was normal. There was no fissuring, erythema, halitosis, or ulceration.

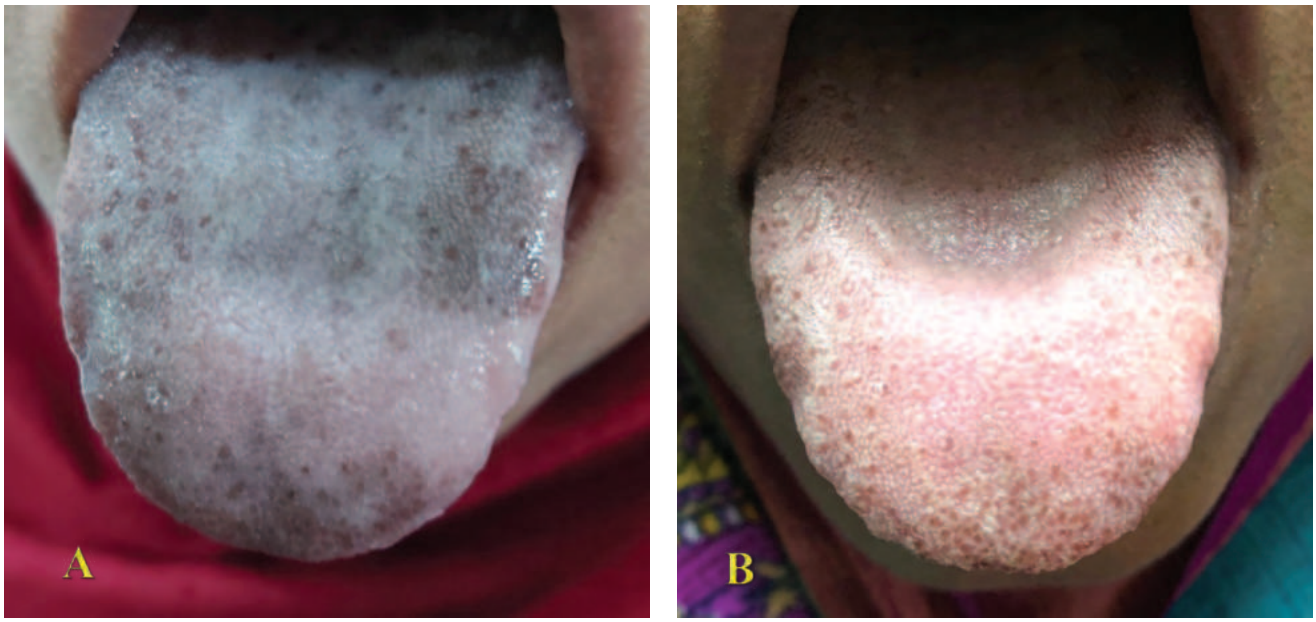

Figure 1: A. Hyperpigmentation in the dorsal surface of the tongue, with asymmetrical black pigmentation along the distribution of the fungiform papillae immediately after radioiodine therapy. B. Improvement of pigmentation towards baseline 3 months following initial presentation.

On scraping the dorsal surface of the tongue with a tooth brush and after repeated mouth washes, pigmentation persisted. The general sensation of the tongue was intact, but a special sensation was lost, except for spicy.

Patient consulted a medicine specialist for the weakness and numbness of limbs and found no definite clue either. Complete blood count (CBC), MRI of the brain and a nerve conduction study (NCS) revealed no abnormality. To see any abnormality in the adrenal glands, ultrasonography of the whole abdomen, including the adrenal glands, was done but all tests were normal.

On post therapy follow-up visits patient developed no new symptoms with gradual disappearance of the darkened areas of tongue and papillae looked almost normal (Figure 1 B) She regained all taste sensations and numbness of the limbs also decreased within 3 to 4 months of post RAIT period.

DISCUSSION

Different types of side effects commonly occur following RAI therapy, such as nausea, dry mouth, pain and swelling of salivary glands, loss of taste sensation, vomiting, and nasolacrimal outflow obstruction. Most of the side effects are transient and result from damage to the salivary glands due to radiation (1-4). This patient presented with black discoloration of the dorsum of the tongue, which is most consistent with post-RAIT. Since the beginning of RAI therapy in 1946, this case indicates one of the rarest side effects following RAI therapy (5). Regarding a mechanism by which RAI could induce pigmentation of the tongue, one study found that the tongue lacks expression of the sodium iodide symporter (6). Though, the absorption of iodide in the tongue tissue is probably not the direct cause of the

impact of RAI. Previous research has demonstrated a higher frequency of fungiform papillae with physiologically enhanced pigmentation (6).

Risk factors for a black, hairy tongue are poor oral hygiene, smoking or alcoholism, excessive drinking of coffee or black tea, immunocompromised conditions (7), and certain drugs such as olanzapine, fluoxetine (8), linezolid (9), penicillin, chloramphenicol, and aureomycin (10). In this patient, no such risk factors were found, as she was in good oral hygiene, non-smoker, non-alcoholic, and had no history of any kind of oral contraceptive, antipsychotic, antidepressant, antibiotic, or any other drug used prior to RAI therapy. Her laboratory investigations reveal no abnormality. Within the next three months of the initial event, improvement of her symptoms occurred despite being on her regular diet, oral hygiene, or without any medication for this symptom.

CONCLUSION

As there are very few documented cases of black pigmentation of the tongue following RAI therapy worldwide till now, including ours, it is an extremely rare side effect following RAI therapy other than the most common side effects, including nausea, dry mouth, sialadenitis, vomiting, and naso-lacrimal duct obstruction. The exact mechanism of this black pigmentation of the tongue following RAI therapy is still not known. We are looking forward to seeking the attention of all other personnel related to the treatment with RAI therapy for such pigmentation of the tongue following RIA as a side effect. The patient can also be informed about this non-harmful, self-limiting side effect, along with other routine advice, before giving radioactive iodine therapy.

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