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Complete Intra-Oral Vitiligo: Report of a Rare Case

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Abstract

Vitiligo is a pigmentary disease causing cosmetic debility. It is defined as an acquired, slowly progressive loss of cutaneous pigment which occurs as irregular, sharply defined patches which may or may not be surrounded by macroscopic hyperpigmentation. It is a disorder that has immense social impact. Both skin and mucous membrane may show depigmentation in vitiligo. Depigmentation in the oral cavity is rarely observed and even more rare is complete intra-oral depigmentation. We present here a case of complete intra oral depigmentation in a known vitiligo patient, which, to the best of our knowledge, is the first of its kind in the English language literature at the time of writing the manuscript. The detailed clinical history, pathogenesis, classification, staging, requirement of biopsy and treatment modalities have been discussed.

Keywords

vitiligo; depigmentation; intra-oral

Introduction

Vitiligo is a puzzling disorder characterized by a disappearance of epidermal and/or follicular melanocytes by mechanisms that are yet not completely understood [1]. Vitiligo is derived from the Latin word 'vitium', meaning a 'defect'. It is a non-contagious acquired pigmentation disorder characterized by well-defined white patches of different shapes and dimensions, progressing with time. It is also called leukopathia, piebaldism or leukoderma [2]. Vitiligo occurs worldwide with a prevalence of 0.5-1%. In India, the incidence of vitiligo is 0.25-2.5% [3]. Although vitiligo is not a life-threatening disease, it has been associated with occurrence of many other autoimmune disorders and concomitant psychosocial difficulties, extensively impacting the quality of life.

Case Report

A 70 year old male patient reported to the Department of Oral Medicine and Radiology which a chief complaint of tooth decay in lower left back tooth region since the last 8-10 months. Medical history was unremarkable and non-contributory to the chief complaint. He gave history of spontaneous depigmentation of the skin of his finger-tips and toes that started approximately 8-10 years back, and had gradually increased in its extent, without any sign of regression. He had visited many physicians with the same complaint and was under medications but there was no evidence of repigmentation or stability of the disease. At the time the patient presented, he was under Ayurvedic medication, which had neither shown any positive result. Family history was negative with respect to the depigmentation disorder.

Upon examination, irregular areas of depigmentation were noted on the dorsal aspects of his extremities involving the skin over the distal phalanges of his left hand, both distal and middle phalanges of his right hand (Fig 1), distal phalanges of his toes, upper and lower lips, (Fig 2) the philtrum region and the skin around the left oral commissure (Fig 3). The areas of depigmentation were non elevated, with no tenderness or secondary changes noted. A thorough general physical examination revealed no depigmented areas elsewhere in the body. Upon intra-oral examination complete loss of pigmentation was noted on bilateral buccal mucosa, vestibules, hard palate, soft palate and marginal, attached and inter-dental gingiva. (Fig 4- 9) The mandibular left first molar tooth demonstrated a deep carious lesion with radiographic pulpal involvement.

Based on the above clinical picture, a clinical diagnosis of oral mucosal depigmentation secondary to generalized vitiligo was arrived at, along with deep dental caries with pulpal involvement in tooth 37. The concerned tooth was appropriately treated by endodontic procedure. The patient was motivated for intra oral biopsy, which he promptly denied as it was not his chief concern and he wished to continue the Ayurvedic medication.

Discussion

Vitiligo is an acquired hypopigmentary disorder having no racial or sexual predilection. It is not only a melanocyte disorder and also involves keratinocytes and Langerhans cells found in the epidermis and follicular epithelium [1].

There are several hypotheses proposed to explain the pathogenesis of vitiligo. The most accepted is the Autoimmune hypothesis, according to which, a variety of almost specific circulating autoantibodies of vitiligo patients are present [4]. They belong to IgG1, IgG2 and IgG3 subclasses and react with multiple antigens not only expressed on pigment cells, but also on other non-pigment cells. But, it is unknown how actually selective destruction of melanocytes occurs in vitiligo, regardless of a heterogeneous antibody response. Literature suggests that melanocytes are much more sensitive to toxic or immune-mediated injury than are other cutaneous cell types. Alternatively, the preferential expression of these antigens on melanocytes may explain the selective destruction [4]. The immunosuppressive effect of certain repigmenting therapies (steroids, UV radiation) indirectly supports the idea of an autoimmune mediated process of depigmentation [4]. The other hypotheses are Neural hypothesis, Self-destructive hypothesis and Biochemical hypothesis [5].

Approximately 30% acquire the disease before the age of 20 years and fewer than 10% develop vitiligo beyond the age of 42 years. Mucosal involvement occurs mostly around body orifices such as the lips, genitals, gingiva, areola and nipples. Involvement of intra-oral mucosa is rare [2]. This case highlights an unusual case of intra oral vitiligo with complete loss of pigmentation of the oral cavity, which, to the best of our knowledge, is the first of its kind in the English language literature at the time of writing the manuscript. Also, the age of onset is around 60 years which makes the case a rarity. In this case, the patient had a dark skin colour, but had score 0 for gingival pigmentation (i.e pink tissue with no pigmentation) according to Dummet-Gupta oral pigmentation index. In a study done by Ponnaiyan et al in 2013, it has been proven that skin colour is correlated to the intensity of mucosal pigmentation which is highly significant and dark skinned individuals have heavy mucosal pigmentation. The present case is an exact opposite to this statement [6]. This again strongly supports the diagnosis of mucosal vitiligo in our case.

Differential diagnosis of vitiligo includes nevus depigmentosus, lichen sclerosus, and chemical leukoderma. Nevus depigmentosus is the most often encountered differential diagnosis. It is a congenital and stable localized leukoderma that has discrete, regular, or often serrated appearance. Vitiligo is acquired and characterized by well circumscribed milky white macules devoid of identifiable melanocytes [2].

Classification

Type of vitiligo [7]	Subtypes
Segmental	Uni, bi or multi segmental, focal, mucosal
Non segmental	Mucosal, generaized, universal, acrofacial
Mixed	According to severity of segmental vitiligo
Non classified	Focal onset, non segmental, asymmetrical, multi focal, mucosal (single site)

Stages

Stages [8]	Extent of depigmentation
0	No depigmentation
1	Partial depigmentation
2	Complete depigmentation
3	Complete depigmenation with > 30% hair whitening
4	Complete hair whitening

Requirement of Biopsy

Biopsy in vitiligo is not recommended to make therapeutic decisions. Digitized photographic assessment is useful for individual lesions, especially when surgery is considered [7]. Surgical modalities of treatment include grafting (autologous), melanocyte transplantation, micropigmentation, dermabrasion, needling, ER-Yag laser, fractional carbon dioxide laser, autologous non-cultured epidermal cell suspension (NCES) etc. Whenever done, histopathologic sections show epithelium devoid of melanocytes and do not show immunoreactivity for HMB-45 antibody.

Treatment Modalities

Treatment for vitiligo can be topical, oral, light and/or surgical therapies [9].

a. Topical therapies include steroids, Vitamin D analogues, Calcineurin inhibitors, Pseudocatalase etc.

b. Oral therapies include psoralen, the immunomodulators -levamisole, Azathioprine; Minocycline, Polypodium Leucotomos,antioxidants etc.

C. Light therapies comprise of PUVA, PUVAsol, UVA : UVA-1,BB-UVA), UVB (BB and NB), Psoralen and Narrow Band-UVB, laser light devices including excimer laser, helium neon laser, non-laser monochromatic excimer light/lamp, sunlight, including special, infra-red light etc.

d. Surgical management as has been discussed in the previous section.

Conclusion

Occasionally, depigmentation in vitiligo can manifest itself first within the oral cavity. In such cases, dental surgeons can detect it early and create awareness in the patient regarding oral depigmentation and the possibility of occurrence in other parts of the body. This can help in early initiation of treatment, thereby combating against a psychosocially distressing disorder in a better way.

The present case is rare owing to the initiation of depigmentation in the subject after the age of 60 years, which occurs in less than 10% of cases of vitiligo. Moreover, complete intra oral depigmentation makes this case an unusual and remarkable one.

Figures

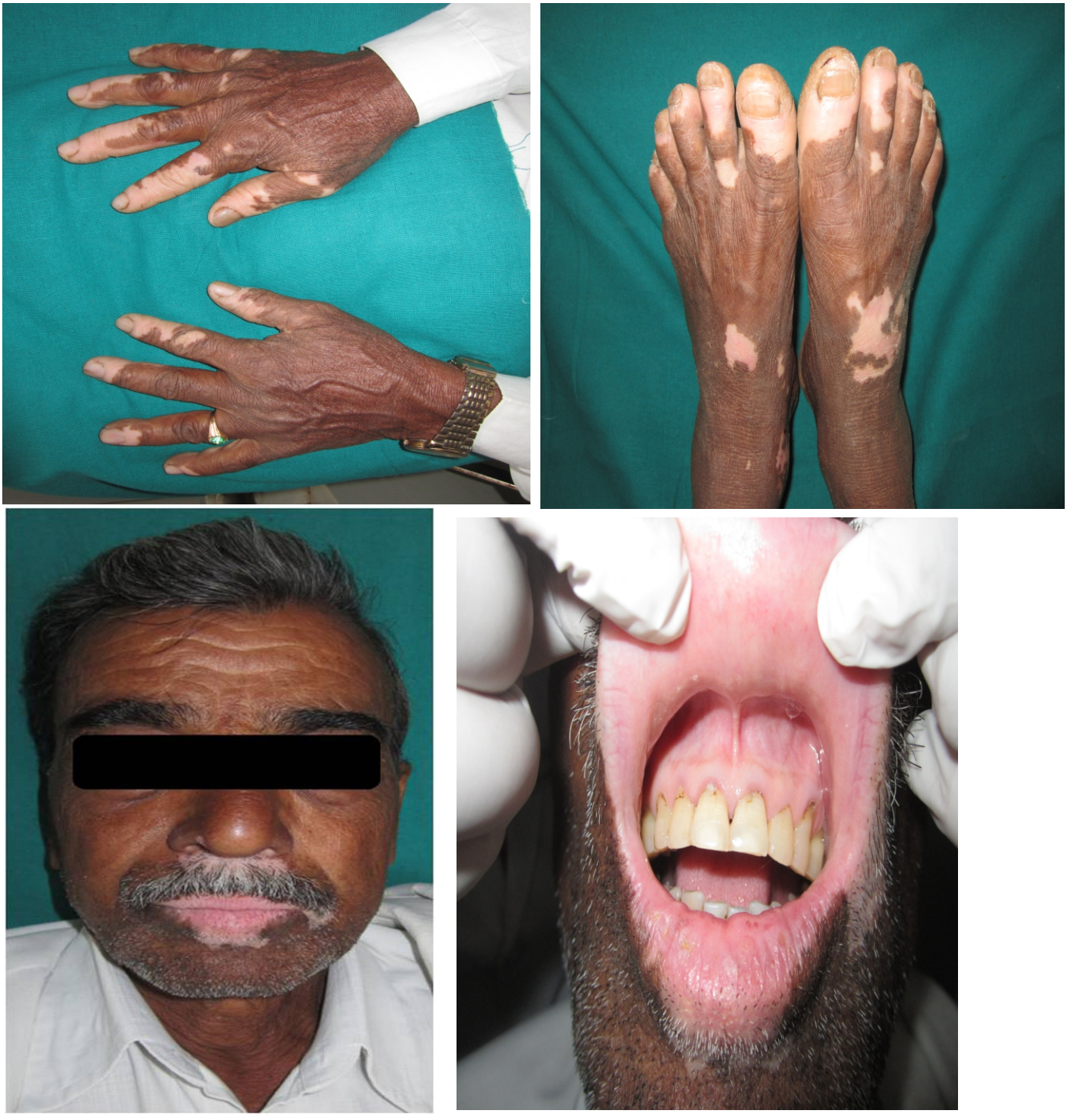

Figure 1,2,3,4

Figure 5,6,7,8,9

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