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Exfoliating White Flakes from the Palate

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Abstract

A 17 year old girl reported with a complaint of white flakes falling off from the mouth since one year and pain along the right side of the face. According to the patient, the frequency of white flakes exfoliating was 150-200 per day which had reduced to 15-20 per day when the patient reported to us. The unusual feature of the present case was the absence of any primary or secondary mucosal lesion or any kind of infection in the oral cavity. The patient was given symptomatic treatment and the phenomenon of exfoliation of white flakes stopped completely within a period of ten months, but the diagnosis of this challenging case could not be met. This paper includes clinical presentation, investigations, and histopathological findings, with special emphasis on the differential diagnosis of this interesting case, which probably could be labeled as one of its kind, and definitely needs documentation.

Keywords: White lesions; Palatal mucosa; Pain; Keratin; Diagnosis; Plantar keratoderma

Case Report

A 17-year-old girl of Asian Indian origin reported to the Oral Medicine, Diagnosis, and Radiology Department, Goa Dental College and Hospital, Bambolim, Goa, India, with a complaint of white flakes falling off from the mouth and pain over the right side of face. According to the patient, the white flakes were pieces of soft tissues in the mouth and were falling off due to accidental cheek biting while eating food. She gave a H/o about 150-200 flakes falling off per day for the past one year, which had decreased to 15-20 flakes per day since the past 4 months. The pain over the right side of face was intermittent, mild, nonradiating, and according to the patient aggravates during menstrual cycle. As per her mother, the patient was a slow learner and had three episodes, over the past 3 years, of talking incoherently for a period of 2-3 minutes followed by temporary amnesia of the incident. The patient had received treatment over the past eight months for the flakes and upper respiratory tract infection, which included antibiotics, vitamins and analgesics. She was put on tab Piracetam 800 mg t.d.s. for 15 days for her problems related to slow learning. Patient was not on any regular medicine for the past 4 months. She had menarche at 13 years with hypomenorrhea and menstrual cycles lasting for 1 to 1$\frac{1}{2}$ days with scanty flow. The patient was the only child, with no siblings. Mother was diabetic and father expired 3 years back due to cardiac arrest. General Examination revealed obesity with body mass index (wt/ht^2): 34.6, (Height: 1.61 mts (5' 4") and Weight: 90 kgs); waist/hip circumference ratio: 0.82, presence of Plantar Keratoderma with normal palmar surfaces (Figure 1) and Hirsutism. Systemic examination included cardio respiratory, CNS and per abdomen examination, which revealed mild Hepatomegaly.

Patient presented with collection of almost 10-12 white flakes in upper right buccal vestibule. Extraoral examination revealed diffuse swelling over right parotid and malar region due to the collection of flakes in the buccal vestibule (Figure 2). Palpation elicited tenderness over right maxilla and along the nasolabial fold. Oropharyngeal examination revealed grade II bilateral tonsillar hypertrophy. For intraoral examination, patient was asked to rinse the mouth with water and was observed for 45 minutes. A white flake appeared over the palate just anterior to the junction of soft and hard palate within a span of 15 minutes (Figure 3). The flake was transparent and firm in consistency. Over a period of next 20 minutes, its size increased and appearance changed from transparent to white opaque resembling a flake of tender coconut (Figures 4a-4c). This flake was easily picked up from the palate with the help of a pair of tweezers. Patient experienced mild pain, but removal of the flakes revealed normal mucosa underneath with no sign of ulceration / erosion or bleeding (Figure 5). Minute observation of palate revealed a shallow depression on the left side of the midline on palate. Oral hygiene was good with extrinsic brown stains present on labial and palatal surface of maxillary teeth. Bidigital palpation of right cheek revealed normal soft tissue texture with a normal appearing Stensen's duct opening with adequate flow of saliva.

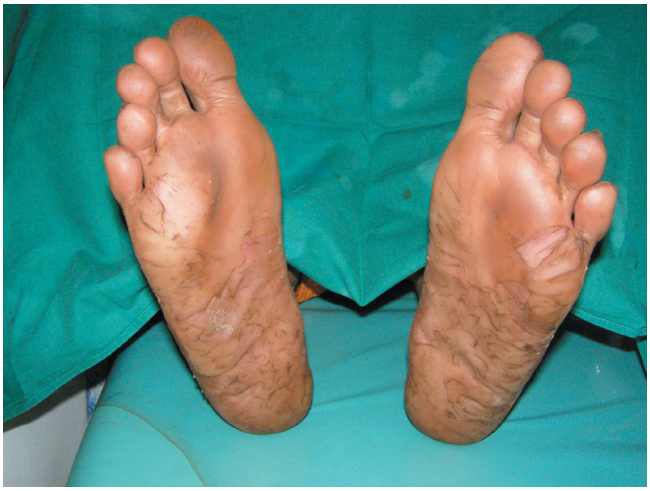

Figure 1: Plantar keratoderma.

Blood investigations revealed eosinophilia (07%) and raised erythrocyte sedimentation rate (ESR) – 42 mm/h. Biochemical investigations, Thyroid function tests, Saliva analysis, Urine analysis,

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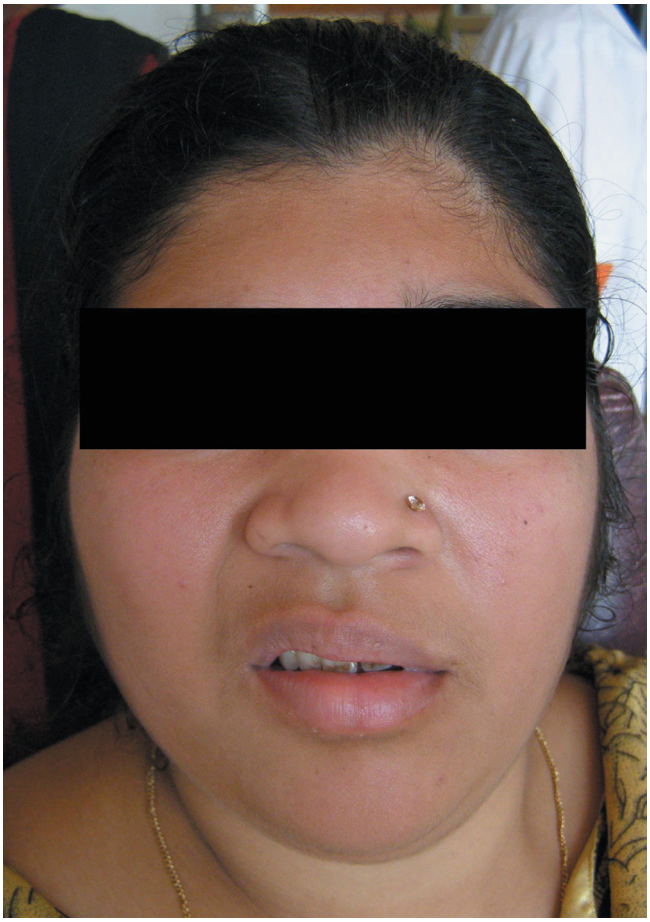

Figure 2: Extraoral appearance showing diffuse swelling over the right parotid and malar region.

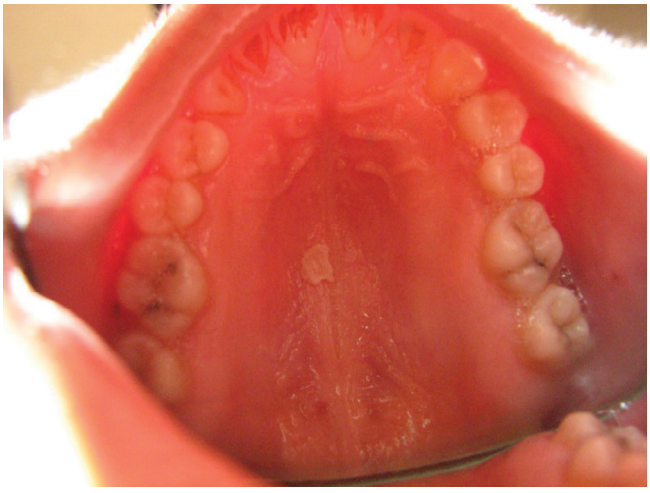

Figure 3: Initial appearance of a white flake on the palate.

and Hormonal assay were noncontributory. Panoramic radiography, PA - soft tissue view with cheek blown for right parotid gland region did not reveal any alteration. Paranasal sinus view showed evidence of mild haziness in right and left maxillary sinus along the lateral walls (Figure 6). Plain CT scan (coronal sections) of the paranasal sinuses

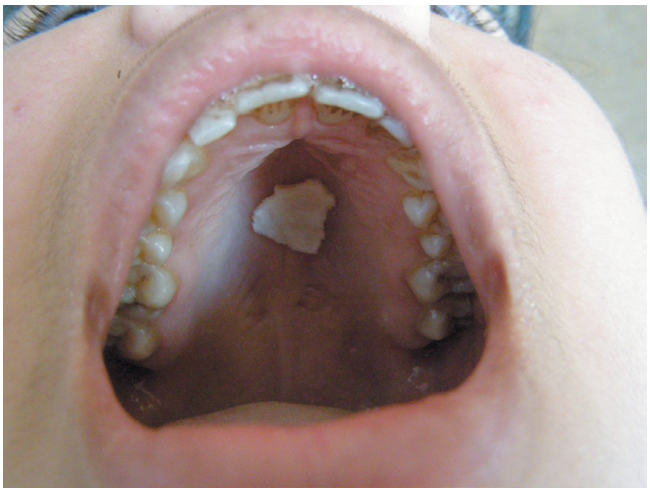

Figure 4a: Appearance of the white flake on the palate after 20 minutes of initial appearance.

Figure 4b: The white flake resembling the flake of tender coconut.

revealed minimal mucosal thickening in the right maxillary sinus with polypoidal mucosal thickening in the left maxillary sinus (Figure 7).

Differential Diagnosis

The positive findings in this case were recurrent white flakes over palate, diffuse tenderness over the right side of face which aggravates during menstruation, hypomenorrhea, hirsutism, obesity, plantar keratoderma, tonsillar hypertrophy, and mild hepatomegaly. The translucent material appearing on the palate could be secretion from the palatal minor salivary glands or it could be the result of hyper-keratinization from palatal mucosa. Other differential diagnosis can include scrapable white lesions of oral cavity, infectious: fungal infection related to maxillary sinus/palate /nasopharynx, gastro intestinal regurgitations, foreign body.

The palatal minor salivary glands are located beneath the epithelium and secrete minor amounts of saliva onto the mucosal surface to keep the mucosa moist. The secretion is purely mucus and

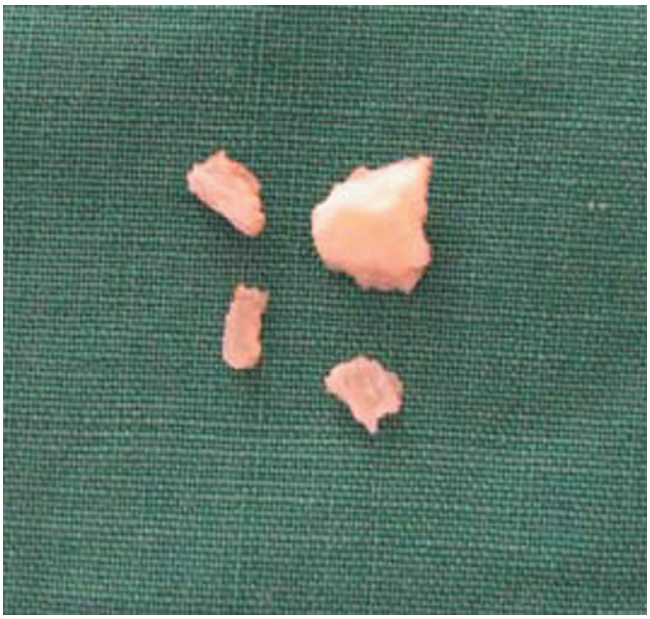

Figure 4c: The white flakes resembling the flakes of tender coconut.

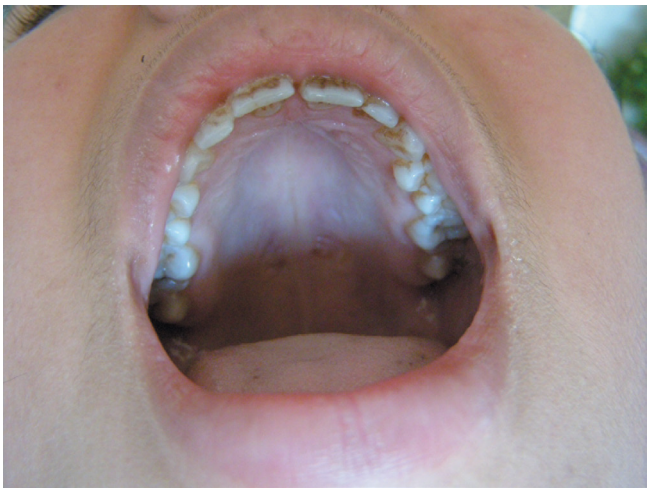

Figure 5: Appearance of the palate; after removal of the flake, with intact mucosal surface.

deposition or crystallization of these secretions is not documented in the dental literature [1]. The scrapable white lesions of oral cavity appear most commonly as plaques. Candidiasis is the most frequently encountered. This can be scraped off the mucosa with a tongue blade leaving a raw, bleeding surface. The pseudomembranous candidiasis presents as soft, white slightly elevated plaques that closely resemble milk curds on an erythematous patch of mucosa. Patient may complain of burning sensation, tenderness or sometimes pain in the affected area [2]. On occasion, oral mucosa that has been crushed by mechanical trauma appears as a sloughing white lesion on the gingivae or other oral sites. History of such a traumatic event is diagnostic, which is absent in the present case. Chronic mild burns usually produce keratotic white lesions whereas more severe burns coagulate the surface of the tissue and produce a diffuse white lesion. If the coagulation is severe, the tissue can be scrapped off, leaving a raw, bleeding, painful surface [3].

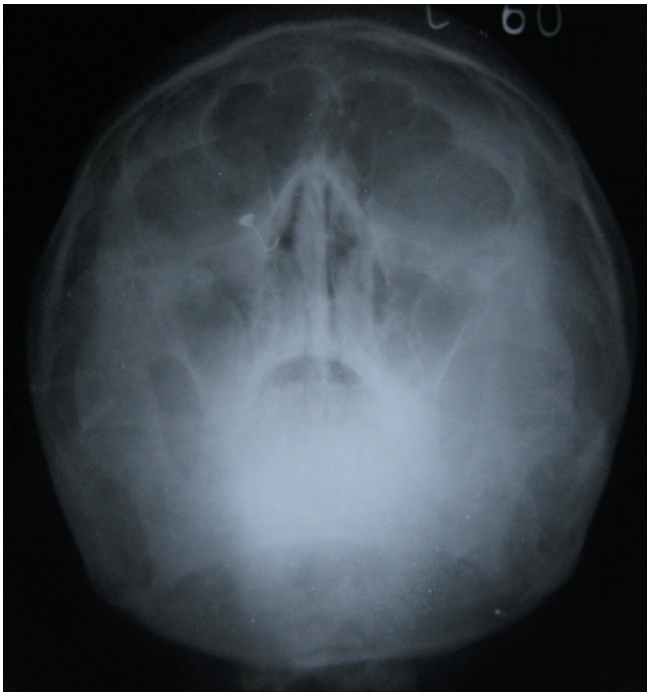

Figure 6: Paranasal sinus view showing mild haziness in right and left maxillary sinus along the lateral walls.

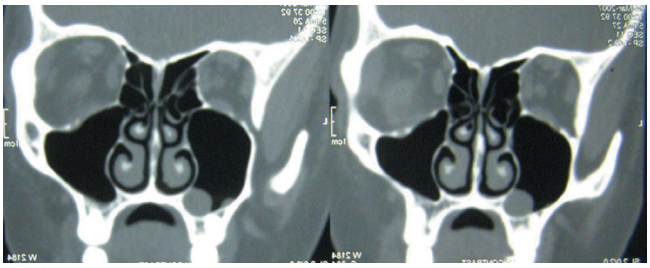

Figure 7: Plain CT paranasal sinuses -Coronal sections showing polypoidal mucosal thickening in the left maxillary sinus.

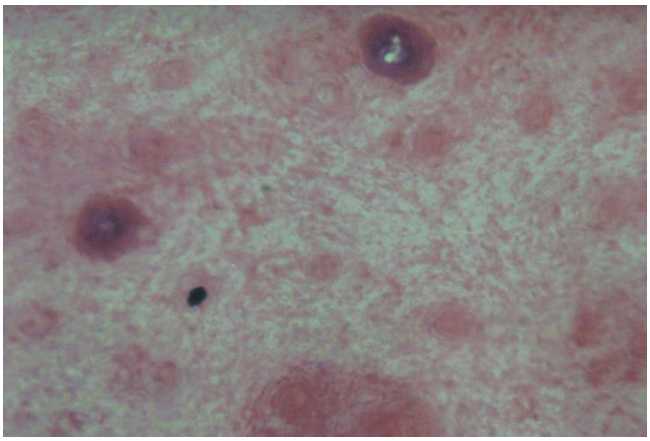

Figure 8a: Histological examination showing normal oral microbial colonies with keratin (hematoxylin-eosin stain).

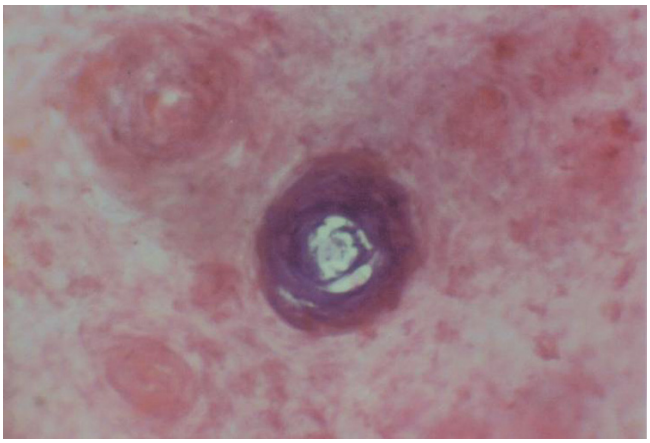

Figure 8b: Histological examination showing normal oral microbial colonies with keratin (hematoxylin-eosin stain, original magnification X40).

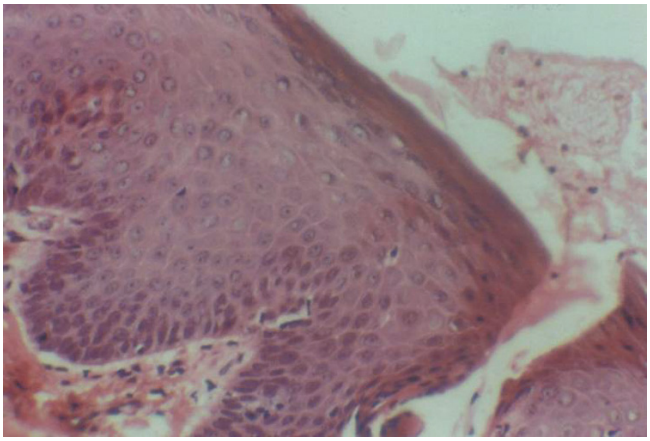

Figure 9: Histological examination showing normal palatal mucosa with stratified squamous orthokeratinized epithelium, fibrovascular connective tissue (hematoxylin-eosin stain, original magnification X40).

The important clinical features of the present case were the rate at which the white flakes were forming and exfoliating from the palatal mucosa, and the absence of any lesion either primary or secondary over the underlying mucosa. Right parotid gland sialadenitis was also included in the diagnosis mainly due to the pain experienced by the patient over the right side of the face. The same was ruled out based on the clinical and radiological examination of the right parotid gland region. Endometriosis is classically defined as ectopic growth of endometrial glands and stroma, [4] possible locations being pelvic cavity, gastrointestinal, urinary tract, pleural cavity, and skin; with the most common symptom being the excruciating pain, at these particular sites. The pain may be experienced constantly, it may be intermittent or it may be related solely to the menstrual period [5]. In a longitudinal study by Leonardo Vecchiet, Maria Adele Giamberardino pressure pain threshold of the masseter and temporalis muscles were assessed across menstrual cycle at four separate sessions during the menstrual, follicular, ovulatory, and luteal phases. They could establish the relationship between muscle pain and fluctuation of the ovarian hormones [6]. The facial pain experienced by the patient could be due to endometriosis.

Management and Diagnosis

The white flakes were sent for culture, and for histopathological evaluation. Culture report was negative for fungal growth. Histological examination showed normal oral microbial colonies with keratin (Figures 8a and 8b). Incisional biopsy of the palate revealed normal palatal mucosa with stratified squamous orthokeratinized epithelium, fibrovascular connective tissue and mucous salivary acini (Figure 9). The patient was put on symptomatic treatment consisting of Analgesics with proteolytic enzyme, (Tab Enzoflam 1 tab t.d.s.); Astringent for local application on the palatal mucosa (Sensoform gum paint: Tannic acid glycerin, Potassium Iodine, Thymol, Menthol) and Tab Bendex (Albendazole) 400 mg H.S. for Eosinophilia. For plantar keratoderma patient was referred to a dermatologist and was advised Cap Vitamin A 25,000 i.u. 1 Cap OD × 30 days and 4% salicylic acid for local application. Patient was further referred to other specialty departments namely ENT, Neurosurgery, General Medicine, Obstetrician and Gynecology. Patient was asked to maintain a chart to record the frequency, duration and progression of the pain over the right side of face to assess its correlation with the four phases of menstrual cycle i.e., menstrual, follicular, ovulatory, and luteal. The pain over the right side of face had an irregular pattern and failed to have any correlation with the menstrual cycle phases. Thus endometriosis was ruled out by the Gynecologist.

During monthly follow up it was noted that the white flakes continued to form at the same region and fall, though the number decreased to 7-8 flakes per day. Repeat plain CT of the paranasal sinuses was performed after five months, which revealed mucosal thickening in both the maxillary sinuses. For further investigation, the paraffin block of the white flakes was submitted to the higher center (Tata memorial hospital, Bombay) for immunohistochemistry for cytokeratin. Microscopic examination revealed hyperkeratotic, parakeratotic, anucleate squamous epithelium with no evidence of malignancy.

The number of white flakes gradually decreased and finally stopped after about 10 months of reporting to the department. The pain over the right side of the face decreased in frequency and severity and gradually stopped. The patient was followed up for next four years then after and was asymptomatic.

Discussion

The scrapable white lesions are most commonly composed of necrotic or coagulated surface epithelium or a mixture of necrotic epithelium, plasma proteins, blood cells, and microorganisms. In case of pseudomembranous candidiasis the plaque is composed of necrotic tissue, pseudohyphae, yeast forms of candida organisms and bacteria [2]. The case presented here had certain unique features, which sets it aside from the usual differential diagnosis for white lesions of oral cavity. These unique features are:

- Sudden onset.
- Rapid production and exfoliation of the white flakes.
- No evident changes on the underlying mucosa of the region of flakes.
- Localized site: palatal mucosa anterior to junction of soft and hard palate.

- Multiple sections of the white flakes showed only increased Keratinous material.
- The Incisional biopsy of the palate failed to show any abnormality at the cellular level or the minor salivary gland acini.

Yosipovitch et al. highlighted in their study an association between obesity and dermatologic conditions. Obesity is implicated in a wide spectrum of dermatologic diseases, including acanthosis nigricans, acrochordons, keratosis pilaris, hyperandrogenism and hirsutism, striae distensae, adiposis dolorosa, and fat redistribution, chronic venous insufficiency, plantar hyperkeratosis etc. The presence of hirsutism and plantar keratoderma could be due to obesity associated changes in skin physiology [7].

The plantar keratoderma is a characteristic feature of certain autosomal dominant inheritant diseases. Jadassohn et al. described a case of fifteen-year-old girl who was admitted to hospital with fungating tuberculosis of the skin. She presented with unusual keratinization of the skin, with extremely thickened nail plates of all the fingers and toes. Abnormalities had been present since birth namely hyperhidrosis of the nose, palms and soles, scanty papular hyperkeratosis of the knees and elbows and a white plaque on the tongue were additional features [8]. Keratosis follicularis, also known as Darier disease (DD) or Darier-White disease, is characterized by greasy hyperkeratotic papules in seborrheic regions such as the forehead, scalp, margin of the scalp, nasolabial folds, ears, chest, and back, nail abnormalities, and mucous membrane changes. Mucosal lesions, most commonly found in the mouth, are detected in approximately 15% of patients, which appear as white papules with a central depression. At times, oral lesions may affect the salivary glands and cause obstruction [9]. The clinical findings of the present case fail to befit in any of the abovementioned autosomal dominant inheritant diseases.

The epithelial lining of the oral mucosa is composed of a constantly renewing cell population. The number of new cells produced is just sufficient to match those lost from the surface due to normal wear and tear [10]. Shklar compared sections of oral mucosa of two age groups of mixed sex a) <16 yrs, b) >60 yrs. The epithelium was thinner and the rete ridges reduced in all parts of the mouth in the older group. The tendency to increased keratinization was detectable only on the hard palate in the later age group [11]. But in the present case increased keratinization was clinically visible in a patient of 17 years of age.

The desmosomal cadherin desmocollin (Dsc1), a transmembrane adhesive protein is expressed in upper epidermis where strong adhesion is required. Dsc1 is part of a desmosomal cell adhesion receptor formed in terminally differentiating keratinocytes of stratified epithelia [12]. Desmoglein 1, also known as Dsg1, is a human gene and is a calcium-binding transmembrane glycoprotein component of desmosomes in vertebrate epithelial cells [13]. Dsc1 and Dsg1 are associated with terminal differentiation and keratinization. Martyn Chidgey et al. performed targeted ablation of the mouse *Dsc1* gene. This resulted in a complex phenotype, showing epidermal fragility together with defects of epidermal barrier and differentiation. The epidermis of neonatal mice showed lesions, which resembled those found in IgA pemphigus, and older mice developed chronic dermatitis. These results demonstrated that Dsc1 contributes substantially to epidermal adhesion and function [14]. The white flake appearing suddenly from the oral mucosa, in the present case could be due to various possible causes as disruption in the transmembrane adhesive proteins causing loss of integrity along the epithelium of the hard palate or an abnormal presentation of certain genetic defect related to Dsg1 gene or a possible hyper keratinization of palatal mucosa. Although, to our knowledge there has not been a single published case report in the dental literature on idiopathic keratinized flakes occurring in the oral cavity, we suggest this case be included among the differential diagnosis of white lesions especially among the scrapable types.

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