Leukoedema of the Tongue in a Renal Failure Patient

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A 20-year-old man with chronic renal failure had a whitish smooth plaque on his tongue. Due to his long standing illness, he had poor oral hygiene. Histopathologic examination showed marked epithelial thickening with parakeratosis and intracellular edema in the prickle cell layer. PAS staining revealed increased amounts of PAS-positive materials in the pale epithelial cells. His tongue lesion persisted during his hospital course.

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Key Words : Leukoedema, Chronic renal failure

Leukoedema is a white or whitish-gray edematous lesion of the buccal and labial oral mucosa.[1] The lesions may be diffuse or patchy, and are usually asymptomatic. Leukoedema may be confused with leukoplakia, Darier's disease, white sponge nevus, pachyonychia congenita, or candidial infection. The condition is seen most commonly among black men, especially with poor hygiene. The histologic appearance simulates that of white sponge nevus.[2]

I herein report a case of leukoedema of the tongue in a 20-year-old chronic renal failure patient.

REPORT OF A CASE

A 20-year-old man was first seen at our hospital with a whitish soft, smooth surfaced plaque on his tongue which was noted 2 weeks ago. He complained of nothing but mild pain when taking food. None of his family members had similar conditions. The area had become more discrete and patchy, and was easily caught between the upper and lower teeth during mastication, and some of the area was slightly tender. The patient did not smoke. He had suffered from chronic renal failure and severe anemia for 3 years, and had been admitted for the management of a recent upper G-I bleeding.

Physical examination revealed a pale and chronic ill-looking patient. His conjunctiva was severely anemic and generalized edema was noted. He complained of right ear otalgia that proved to be otitis externa. He had a discrete, 1 ×5㎝ sized, whitish, non-tender, velvety plaque along the margin of his tongue(Fig. 1). Some areas showed irregular, fine, discrete erosions with a "moth-eaten" surface. Similar lesions were not found on any other part of the mucosa. The plaque could not be removed with a wooden tongue blade.

A 3㎜ punch biopsy specimen was taken at the margin of the plaque. Histologically, the epithelium was pale and showed severe acanthosis and parakeratosis with marked intracellular

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edema(Fig. 2, 3). Vacuolation of epithelial cells was common, and a granular layer was lacking. There was no dermal inflammation or atypical cells. Staining with PAS showed increased PAS-positive materials in the pale epithelial cells(Fig. 4).

Laboratory work revealed severe anemia(Hb 8.9g/dℓ) with leukocytosis(WBC 10,800), albumin and glucose in the urine, and other signs of azotemia(calcium 7.1㎎/dℓ, phosphorous 11.2㎎/dℓ, BUN 173㎎/㎖, creatinine 30.1㎎/㎖, uric acid 14.4㎎/dℓ). His laboratory abnormalities resolved after several blood transfusion and emergency hemodialysis, but his tongue lesion persisted, even after oral gargling with chlorohexidine and lidocaine. He was discharged and did not return for further mangement, so no specific therapy was initiated.

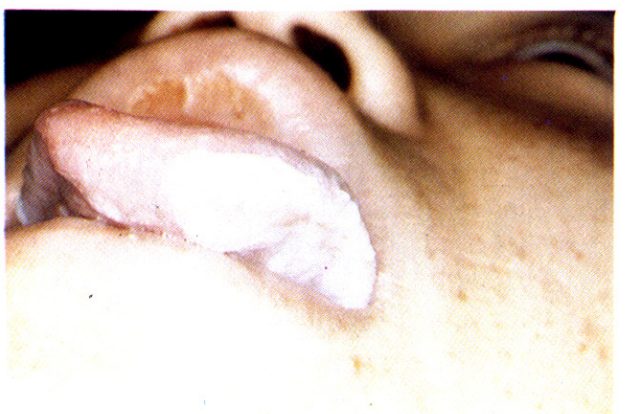

Fig. 1. A discrete, 1×5㎝ sized, whitish, non-tender, velvety plaque along the margin of the tongue.

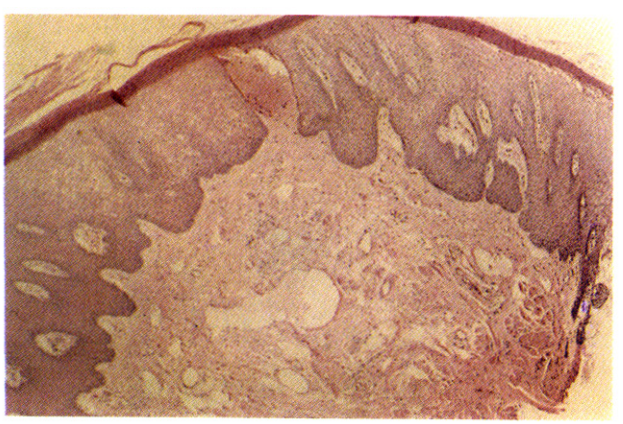

Fig. 2. Pale epithelium showing severe acanthosis and parkeratosis with marked intracellular edema(H & E stain, ×40).

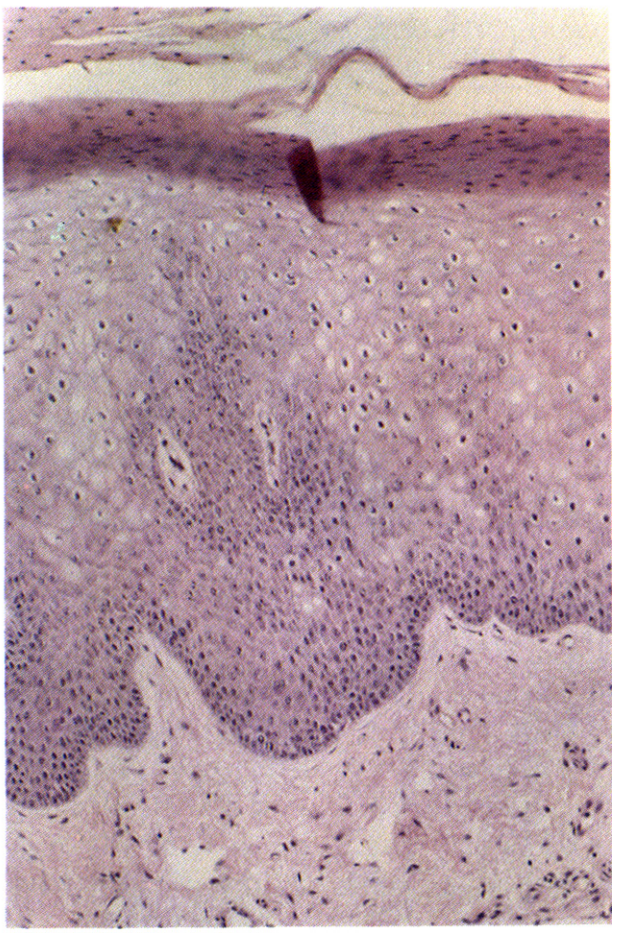

Fig. 3. Vacuolation of epithelial cells was common. There was no dermal inflammation or atypical cells(H & E stain, ×100).

DISCUSSION

Leukoedema of the oral mucosa is a relatively frequent condition.[2] In 1953, Sandstead and Lowe[1] described, in a survey of 646 psychiatric patients, an asymptomatic condition of the buccal mucosa which was present in 43% of the white patients and 90% of the black patients

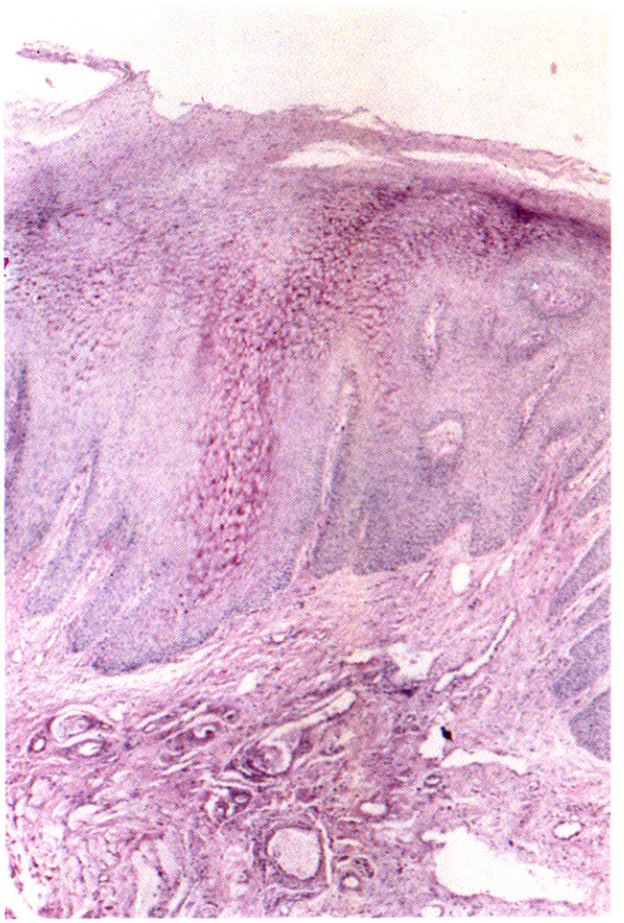

Fig. 4. PAS staining showed increased PAS-positive materials in the pale epithelial cells(PAS stain, ×100).

A diffuse mild involvement was the most common clinical form, and in more advanced cases, impressions of the teeth were prominent along the occlusal line, and desquamation occurred, leaving a "moth-eaten" eroded surface. The entire buccal mucosa might be involved, although the middle and posterior portions of the cheek were principally affected. The lesions were usually bilateral, and the oral surface of the lips was frequently involved. Men were affected more frequently than women. There was a strong relationship to poor oral hygiene, although factors such as age, tobacco habits, number of dental fillings, and positive findings for syphilis appeared to be unrelated. Histologically, the lesions showed epithelial thickening, intracellular edema of the Malphigian layer, parakeratosis without an increase in keratohyaline granules, and broad, irregular rete ridges. They named the condition "leukoedema". Martin and Crump[3] later studied the condition among 1,000 black children and youths and found an incidence of 50.8%. They found that the severity increased with age, and they demonstrated a definite relationship to poor oral hygiene. They found no sex predilection and no familial correlation. Durocher et al.[4] reported a much higher incidence(97%). They proposed that the condition be considered a variation of normal. More recent studies show different rates of prevalence among races, such as 49.07 %(Swedish),[5] 24.4%(South Africans),[6] 12.4% (Thailand),[7] and even 0.03%(white Americans).[8] Some authors proved strong relationship between prevalence rates of leukoedema and smoking habits.[6,7,9]

Microscopic observations[10] revealed that the clinical appearance of the diffuse gray-white lesions was due chiefly to a retained superficial layer of parakeratotic cells and that "edema" of the cells in the oral epithelium(particularly in the prickle cell layer) is so frequent as to have no apparent histologic significance.

Several mucosal entities that may appear as white plaques should be differentiated from leukoedema.[2] The most frequent misdiagnosis is probably that of leukoplakia. Clinically, the white plaques of leukoedema are more edematous than those of leukokeratosis or leukoplakia. Histopathologically leukoedema demonstrates a marked intracellular edema of the Malphigian cells with acanthosis, which is much less prominent in leukokeratosis or leukoplakia. There is no inflammatory cell infiltration in the upper corium, as is commonly seen in leukoplakia. Leukoedema also does not show dyskeratotic epithelial cells, a feature of leukoplakia. White sponge nevus is a dominantly inherited condition that may be present at birth or may appear in childhood.[11] Extensive areas of the oral mucosa may be involved with a thickened,

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folded, creamy-white plaque. The rectal mucosa, the vagina, the nasal mucosa, and the esophagus sometimes show similar changes. Electron microscopic studies of white sponge nevus demonstrated disturbances of the tonofilament system and, in distinction to normal buccal cells, numerous Odland bodies. In an ultrastructural study of leukoedema, Duperrat et al.[12] found an abrupt absence of the tonofilaments in the suprabasilar area, and the presence of prominent perinuclear vacuoles. Later Van Wyk et al[13] showed aggregates of ribosomes and electron dense masses, or a network of electron-dense material, or an electron-dense network with a central dense core, or a solid body with a few central cavities. Histochemical stainings and RNA extraction studies showed that they are probably abnormal forms of keratohyaline granules and that ribosomes are an important component of their composition. The clinical lesions and the histopathologic features of white sponge nevus and leukoedema are very similar. However, in addition to their autosomal dominant mode of inheritance, white sponge nevus lesions remain unchanged, while leukoedema may have exacerbations and remissions; white sponge nevus also may involve the genital or anal region, which is very unusual for leukoedema.[2] Other hereditary disorders such as dyskeratosis congenita, pachyonychia congenita, Darier's disease, and hereditary benign intraepithelial dyskeratosis can be readily differentiated by not only hereditary factors but other clinical manifestations.[2] Infectious diseases can be easily differentiated from leukoedema by culture or histopathologic examinations of the oral lesions.[2] Other acquired disorders, including palatitis nicotina(smoker's palate), oral florid papillomatosis, squamous cell carcinoma, and lesions secondary to cheek biting, must be ruled out.

In conclusion, the dermatologist should be aware of this frequent but poorly recognized entity of the oral mucosa. Various white keratotic lesions of the oral mucosa need to be differentiated from leukoedema. This condition is benign, and generally asymptomatic, but in severe case, it can often be associated with pain or a burning sensation, requiring a biopsy specimen for differentiation from more important diseases.

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