A Case of White Sponge Nevus-like Lesion of the Oral Mucosa Successfully Treated with Azithromycin.

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A 30-year-old Japanese man presented white spongy plaques on his buccal mucosa and lower lip. The surface of the lesion showed irregular and small portions of the plaques were desquamative. Histopathologically, stratified squamous epithelium characterized by acanthosis and parakeratosis of surface layer was identified. Marked intracellular edema without spongiosis and vacuolated spinous cells were observed. Topical steroid and laser treatment were not effective, but the lesion disappeared completely after treatment with azithromycin.

Key words: white lesion of the oral mucosa, white sponge nevus, antibiotics.

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Introduction

Many lesions of the oral mucosa may appear in the form of white patches. Among them, white sponge nevus (WSN) is a rare autosomal dominant disorder of non-cornifying squamous epithelium and is characterized widespread white, soft, thick plaques of the oral mucosa (1). Histologically, the epithelium is hyperplastic and has a thick, irregular, parakeratotic layer which is grossly edematous and has a so-called basket-weave appearance. Generally, these lesions are asymptomatic. Many treatments, including vitamins, antihistamines, nystatin, and liquid nitrogen have been tried but are unsuccessful (1), so the treatment of WSN is considered unrewarding. However, recent reports have indicated the efficacy of penicillin (2) or tetracycline (3, 4) on WSN lesion of the oral mucosa. We report a case of WSN-like white lesion of the oral mucosa which completely disappeared after treatment with azithromycin.

Case Report

A 30-year-old Japanese man visited the Department of Dentistry, Oral and Maxillofacial Surgery, Jichi Medical School Hospital complaining of a sore on his lower lip and inner cheek. The patient first noticed the white lesion about 5 years earlier and visited a local hospital, but there was no improvement of the lesion. He experienced discomfort and roughness in his oral cavity. Oral examination revealed extensive white spongy plaques on his buccal mucosa and lower lip (Fig. 1). The surface of the lesion showed irregular and small portions of the plaques were desquamative. The patient had no family history and the affected area was limited to the oral mucosa. He had a history of smoking for several years but no history of any trauma to the oral cavity. Candida was not isolated from the lesion. We performed biopsy of the lip lesion. After the biopsy, the patient noticed some improvement in the oral discomfort. The relief was immediate after medication of antibiotics (cefcapene pivoxil 300 mg/day, 3 days), and it lasted several days. Histopathologically, stratified squamous epithelium characterized by acanthosis and parakeratosis of surface layer was identified (Fig. 2). Marked intracellular edema without spongiosis was observed in the spinous cell layer; however, the basal and parabasal layers were unremarkable. Vacuolated spinous cells had abnormal cell membranes and pyknotic nuclei in the center of cells. No apparent atypical features were noted and inflammatory infiltration was absent.

We tried to treat the lesion with contact laser (Nd: YAG Contact Laser, wavelength: 1064 nm, 10 W). However, the treatment was not effective at all, so we gave antibiotics azithromycin (500 mg/day, 3 days). The symp-
tomatic relief was again dramatic and the lesion completely disappeared (Fig. 4). There has been no recurrence for 1 year.

Discussion

There exist a number of white lesions of the oral mucosa. Some are caused by local factors, but others may be associated with systemic diseases. White sponge nevus (WSN) is an uncommon autosomal dominant disorder (1). Clinically, the lesions are white, nonpainful, spongy plaques of various size, the extent of which may vary from time to time. The surface of the plaques may peel away from the underlying tissue. The nasal, esophageal, laryngeal, vaginal, and anal mucosa have been reported to be affected (1). Nonfamilial cases have been also reported and usually these had more extensive plaques (4). Our case had no family history and the affected area was limited to the oral mucosa.

The pathogenesis of WSN has been suggested to be an alteration in the distribution of tonofilaments in the epithelial cells. Recent reports indicated point muta-
tions in keratin 4 and keratin 13 genes (5). Another report concluded that the thickening of superficial layer is due to shedding disturbance relating to dysfunction of Odland bodies (6). However, the exact pathogenesis of WSN is unclear.

Because the lesions are usually asymptomatic, treatments may not be necessary in such case (1). On the other hand, there are some reports on the therapeutic trials of WSN. Although vitamin A, antihistamines, nystatin and liquid nitrogen were generally unsuccessful, parenteral penicillin, and topical tretinoin have used with varying success (2, 3). Lim et al. (4) reported the case who did not respond penicillin injection, but the symptoms disappeared and the lesion improved after treatment with 0.25% tetracycline oral rinse.

In the present case, the symptoms temporally improved after the biopsy and medication of antibiotics (cefcapene pivoxil), and the symptomatic relief was completed after treatment with azithromycin.

On the pharmacological mechanisms, some local antimicrobial effects may be considered. McDonagh et al. suggested that the epithelial abnormality of WSN promotes growth of organisms not usually pathogenic, which might account for the usefulness of antibiotics (3). As exact mechanism of antibiotics on WSN are not clear and symptomatic relapse was reported (2), a long-term follow up must be necessary for the patient. Although our case had no family history, the affected area was limited to the oral mucosa and the histopathological findings was not typical, the white lesion was most suspected of an incomplete type of WSN based on the clinical features, course and histopathology.

References

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