GRINSPAN’S SYNDROME - A CASE REPORT

Halimi S1*, Ferizi M1, Gerqari A1, Krasniqi N2, Ferizi M2

1. Department of Dermatovenerology, University Clinical Centre of Kosovo, Pristina, Republic of Kosovo
2. University of Prishtina, Medical Faculty, Pristina, Republic of Kosovo

Correspondence
Dr Sadije Halimi . Department of Dermatovenerology, University Clinical Centre of Kosovo, Pristina, Republic of Kosovo
Email: sadijehalimi@gmail.com


ABSTRACT

Some internal disease can have impact in the skin. Grinspan syndrome is a syndrome characterized by presence of the triad: hypertension, diabetes mellitus and oral lichen planus. Grinspan in 1963 reported 23 patient having oral erosive lichen planus, associated with diabetes mellitus. In 1965 this triad was subsequently referred to as “Grinspan syndrome” by Grupper and Avul. Oral lichen planus is thought to be a result of the drugs used for treatment of hypertension and diabetes mellitus because drug therapy for diabetes mellitus and hypertension is capable of producing lichenoid reactions of the oral mucosa but this is not confirmed. This paper presents a case of mucocutaneous lichen planus associated with diabetes mellitus, and vascular hypertension. Our case is a 63 years old female. During intraoral examination, we can see white and ulcerated lesions all over the oral mucosa and pain during mastication, upon for the past one year, associated with diabetes mellitus and arterial hypertension. Some internal disease can have impact in the skin. Grinspan syndrome is characterized by presence of the triad: hypertension, diabetes mellitus and oral lichen planus. The patients with oral lichen planus, should have a healthy lifestyle, well-balanced diet and stress reduction because this disease can be controlled but not eliminated.

Keywords: Lichen planus, internal diseases, Grinspan's syndrome

INTRODUCTION

Lichen planus is mucocutaneous inflammatory disease that affects skin, mouth and genital region. The disease mainly occurs in females, in middle aged and elderly people. Oral lichen planus in children is rare. The prevalence in the general population is about 1.27-2.0%1-4. Oral lichen planus is chronic inflammatory condition that affects mucous membranes inside mouth and can appear as white, lacy patches, red, swollen tissues, or open sores. These lesions may cause burning, pain or other discomforted.
The etiology of oral lichen planus is unknown, but genetics and immunity may be involved. It is thought to be a T cell mediated autoimmune reaction. This autoimmune process triggers apoptosis of the cells. Several cytokines are involved in lichen planus, including tumor necrosis factor alpha, interferon gamma, interleukin-1 alpha, interleukin 6, and interleukin 8. This autoimmune, T cell mediated, process is thought to be in response to some antigenic change in the oral mucosa, but a specific antigen has not been identified. If a triggering agent is identified, this is called a lichenoid reaction not a lichen planus. These may include a reaction to some medicines, such as: diuretics, spironolactone, antimalarials, gold salts, Metformin and penicillamine. Reactions to amalgam (metal alloys) fillings can cause the lichenoid reactions too. Lichen planus may be caused by stress, that is often mentioned in a patient's anamnesis.

Grinspan found association of oral lichen planus with diabetes mellitus and vascular hypertension for this time called Grinspans’ syndrome. Other reported Grinspan joint with dyslipidemia and chronic liver disease.

CASE REPORT

A. Case History

A 63 year old female Albanian patient (H.M.) was referred to dermatologist from dentist with a complaint of mucocutaneous signs, with oral whitish freckles, polygonal papulous, pruritic pigmented lesions of varying size 1-5mm all over the body but concentrated more on the abdomen, flexor surfaces of forearm and legs with symmetrical distribution. During intraoral examination, we can see leucoplaziform and ulcerated lesions all over the oral mucosa and pain during mastication, upon for the past one year. Over the dorsal surface of the tongue were seen lacy reticular patches and streaks.

The patient had been diagnosed with diabetes mellitus in 1999 under medication with Glibenclamide (diabos) - 2 x 5 mg daily, and vascular hypertension in 2005 being treated with Scopryl tablet 20mg - 2 x ½ daily, Amlodipine tablet 5 mg.1 x 1 at middle of the day. She confirmed that similar mucocutaneous changes, diabetes and arterial hypertension together have in other family members. The patient had been operated from cholecystectomy before 30 years.

B. Laboratory investigations

SE = 43, RBC = 4.25X1012/l, MCV = 84.4fl, HCT = 35.9, PLT = 138, MPV = 8.9, PDV = 12.6, PCT = 0.12, LPCR = 21.1, WBC = 15.7x109 /l HGB = 13.5g/dl, MCH=31.8pg, MCHC = 37.6g/dl, lym = 4.0x109/l, gran = 10.6x109/l, mid = 1.1x109/l, Glycosae = 15.89mmol/I, Cholesterol = 8.62mmol/I, Triglyceride = 1.9mmol/I, Urea = 7.71mmol/I, Creatinine = 75.4umol/I, T Bilirubin = 5.5 umol/I Albumin = 43.8g/I, ALT/GPT LIQ=20 U/l, AST/GOT LIQ = 20 U/l, CRP-HL = 9.2 mg/L
On the forearms they are apparent bumps that are shiny, firm, and reddish purple. Sometimes the bumps have tiny white lines running through them.
C. Abdominal Echo

Liver slight enlarged with 15.4 DLMCL, slight hyperechogenic parenchym, the kidney with calcifications. Organs and other structures without pathological signs.

D. Histological examination of biopsy

Histological examination of biopsy from the lesion oral mucosa, shows atrophic squamous epithelium, extensive ulceration and same area replace by granulation.

E. Treatment

For oral lichen planus the patient was treated with topical application of Tacrolimus ointment 0.1% 1 - 2 applications daily and hyalurinic acid spray (Ialoclean spray), 4 - 5 applications daily after meals. After 4 weeks with this treatment the evolution of oral lesions was favorable with partial epithelisation and improvement of functional
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symptoms. The patient needs to manage disease by monitoring and repeat oral lesions biopsy, to prevent malignant transformation to an invasive squamous cell carcinoma.

DISCUSSION

Lichen planus is a noninfectious, pruritic papular skin disease, commonly affecting mucous membrane characteristic by the appearance of characteristic smooth, flat, reddish-blue, polygonal papules, often with whitish freckles (Wickham'Striae). Oral lichen planus may affects patients with skin lichen planus but may occur without skin lesions elsewhere. It usually affects females older than 45 years, but it can occur at any age.

The association of erosive oral lichen planus with diabetes mellitus and arterial hypertension was first reported by Grinspan in 1963.

The etiology of oral lichen planus is unknown but genetics and immunity may be involved.

The hypothesis of an iatrogenic origin of the oral lesions has also been stated, considering the inherent multiple drug association due to the two chronic co morbidities with a lethal risk.

The treatment to diabetes mellitus and arterial hypertension responds to antigens in the oral epithelium. The most common drugs which may cause lichenoid reactions are: diuretics, gold salts, beta blockers, antimalarials, metformin and penicillamine and reactions to amalgam (metal alloys) fillings (or when they are removed/replaced). Lichen planus has been associated with chronic liver disease, primary biliary cirrhosis, hepatitis B and C, diabetes mellitus hypertension and other disease.

Skin lesions can be seen in diabetes mellitus according to metabolic disorder of glucose, insulin and lipids.

In our case the patient was treated for oral lichen earlier by other dentists but after laboratory and endocrinologist, cardiologist and dermatologist investigations it was established that patient has suffering from Grinspan syndrome. The patient was treated with medications for diabetes and arterial hypertension. For oral lichen planus the patient was treated under topical application of Tacrolimus ointment 0.1%, 1 - 2 applications daily and hyalurinic acid spray (Ialoclean spray) 4 - 5 applications daily after meals. Four weeks after treatment the evolution of oral lesions results with partial epithelisation and functional improvement.

CONCLUSION

Some internal disease can have impact in the skin.

Oral Lichen planus is a chronic inflammatory disease with an immune substrate that may associated with a cardiovascular, endocrine, liver and other disease.

Grinspan syndrome is characterized by presence of the triad: vascular hypertension, diabetes mellitus and oral lichen planus. Our case of Grinspan’s syndrome associates diabetes mellitus with high levels of glucose, dyslipidemia with high levels of cholesterol and vascular hypertension.

The patients with oral lichen planus, should have a healthy lifestyle, well-balanced diet and stress reduction because this disease can be controlled but not eliminated.
They need regular monitoring and repeat oral lesions biopsy because they may be at risk of developing mouth cancer in the affected areas.

AUTHOR'S CONTRIBUTIONS

SH, MF, AG, for analyzed and interpreted the patient's data. SH was a major contributor in writing the manuscript. All authors read and approved the final manuscript. NK and MF contributed in collaborations with Stomatology and Pathology Departments.

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CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

COMPETING INTERESTS

There is no conflict of interest.

REFERENCES