

Clinical manifestations of Grinspan–Potekaev syndrome on the buccal mucous membrane (a case report)

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A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation; D – writing the article; E – critical revision of the article; F – final approval of the article

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Grinspan–Potekaev syndrome is a rare disorder facing dentists in practical activity. That is why the detailed analysis of the disease clinical course peculiarities is important and urgently needed.

The aim of the work: to analyze the clinical course of Grinspan–Potekaev syndrome on the buccal mucous membrane using the example of a patient treatment in the dental clinic Denta Plus (Zhytomyr city).

Materials and methods. Biblisemantic method (analysis, correlation, comparison, generalization, systematization of scientific literature on the studied issues), system analysis and logical generalization (for developing the algorithm of diagnostics and treatment), medical documentation analysis of the patient, who sought medical assistance in the dental clinic Denta Plus, were used.

Results. The clinical course of Grinspan–Potekaev syndrome on the buccal mucous membrane was described and analyzed. The conducted diagnostic methods allowed to determine the diagnosis, to develop and to apply the complex drug therapy that positively influenced the general well-being, promoted the transformation of erosive-ulcerous form of lichen planus into less aggressive and safer exudative-hyperaemic one. The difficulties during the diagnostic and differential diagnostic stages concerned the fact that typical skin lesions were not observed and the “fern pattern” on the buccal mucous membrane (within the period of exacerbation) was not clear. Besides, the patient did not have any complaints specific to diabetes mellitus, therefore a blood glucose level and glycemic profile were determined only in the process of case follow-up, the patient was further referred to an endocrinologist, who diagnosed type 2 diabetes mellitus.

Conclusions. The described case of Grinspan–Potekaev syndrome allowed to analyze the clinical course of the disease in the oral cavity within the exacerbation period and to examine the components of diagnostic and treatment process. The treatment response in Grinspan–Potekaev syndrome (with isolated manifestations of lichen planus on the buccal mucous membrane) depends not only on a dentist competence but also on cooperation between other specialists: dermatologist, oncologist, psychotherapist, family physician, and endocrinologist. Despite the positive clinical effect, the need to follow-up the further course of the disease is associated with the possibility of erosive-ulcerous form of lichen planus relapses and its malignant transformation.

Ключові слова:

синдром
Гриншпана–
Потекаєва,
порожнина
ротова, ознаки та
симптоми.

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Клінічні прояви синдрому Гриншпана–Потекаєва на слизовій оболонці порожнини рота (клінічний випадок)

В. М. Косенко, І. М. Скиба

Синдром Гриншпана–Потекаєва – рідкісна патологія, яку лікарі-стоматологи виявляють під час практичної діяльності. Важливий і вкрай необхідний детальний аналіз особливостей клінічного перебігу цього захворювання.

Мета роботи – проаналізувати клінічний перебіг синдрому Гриншпана–Потекаєва на слизовій оболонці порожнини рота на прикладі лікування пацієнтки, яка звернулася в стоматологічну клініку Дента Плюс (м. Житомир).

Матеріали та методи. Під час роботи використали такі методи дослідження: бібліосемантичний (аналіз, зіставлення, порівняння, узагальнення, систематизація відомостей наукової літератури), системного аналізу та логічного узагальнення (для складання алгоритму діагностики та лікування), аналізу медичної документації пацієнтки, яка звернулася у стоматологічну клініку.

Результати. Описано та проаналізовано клінічний перебіг синдрому Гриншпана–Потекаєва на слизовій оболонці порожнини рота. Діагностичні заходи дали підстави встановити діагноз та розробити, застосовувати комплексну медикаментозну терапію, що позитивно вплинула на загальне самопочуття, сприяла поліпшенню стану пацієнтки – переходу ерозивно-виразкової форми червоного плаского лишая в менш агресивну та безпечну ексудативно-гіперемічну. Труднощі на етапах діагностики та диференційної діагностики були пов'язані з тим, що не фіксували типові ураження шкіри, а «симптом папороті» на слизовій оболонці порожнини рота (в період загострення) був нечітко вираженим. Крім того, скарг, що характерні для цукрового діабету, в пацієнтки не було, тому тільки у процесі динамічного спостереження здійснили дослідження крові на цукор, встановили глікемічний профіль. Пацієнтку направили до лікаря-ендокринолога, який діагнував цукровий діабет 2 типу.

Висновки. Наведений випадок синдрому Гриншпана–Потекаєва дав можливість проаналізувати клінічні прояви хвороби в порожнині рота в період загострення, вивчити складові лікувально-діагностичного процесу. Ефект лікування синдрому Гриншпана–Потекаєва (з ізольованими проявами червоного плаского лишая на слизовій порожнині рота) залежить не тільки від ерудиції стоматолога, але й від його вміння співпрацювати з фахівцями різних спеціальностей: дерматологом, онкологом, психотерапевтом, сімейним лікарем, ендокринологом. Незважаючи на позитивний клінічний ефект, необхідність динамічного спостереження за перебігом захворювання зумовлена можливістю рецидивів ерозивно-виразкової форми червоного плаского лишая та його злоякісної трансформації.

Клинические проявления синдрома Гриншпана–Потекаева на слизистой оболочке полости рта (клинический случай)

В. Н. Косенко, И. Н. Скиба

Синдром Гриншпана–Потекаева – редкая патология, с которой сталкиваются в практической деятельности врачи-стоматологи. Важен и крайне необходим детальный анализ особенностей клинического течения этой болезни.

Цель работы – проанализировать клиническое течение синдрома Гриншпана–Потекаева на слизистой оболочке полости рта на примере лечения пациентки, которая обратилась в стоматологическую клинику Дента Плюс (г. Житомир, Украина).

Материалы и методы. В ходе работы использованы такие методы исследования: библиосемантический (анализ, сопоставление, сравнение, обобщение, систематизация данных научной литературы), системного анализа и логического обобщения (для составления алгоритма диагностики и лечения), анализ медицинской документации пациентки, которая обратилась в стоматологическую клинику.

Результаты. Описано и проанализировано клиническое течение синдрома Гриншпана–Потекаева на слизистой оболочке полости рта. Диагностические мероприятия позволили установить диагноз, разработать и применить комплексную медикаментозную терапию, которая положительно повлияла на общее самочувствие, способствовала переходу эрозивно-язвенной формы красного плоского лишая в менее агрессивную и безопасную экссудативно-гиперемическую. Трудности на этапах диагностики и дифференциальной диагностики были связаны с тем, что не фиксировали типичные поражения кожи, а «симптом папоротника» на слизистой оболочке полости рта (в период обострения) был нечётко выражен. Кроме того, жалоб, характерных для сахарного диабета, у пациентки не было, поэтому только в процессе динамического наблюдения проведены исследования крови на сахар, установлен гликемический профиль. Пациентка была направлена к врачу-эндокринологу, который диагностировал сахарный диабет 2 типа.

Выводы. Описанный случай синдрома Гриншпана–Потекаева позволил проанализировать клинические проявления болезни в полости рта в период обострения и рассмотреть составляющие лечебно-диагностического процесса. Эффект лечения синдрома Гриншпана–Потекаева (с изолированными проявлениями красного плоского лишая на слизистой полости рта) зависит не только от эрудиции стоматолога, но и от его умения сотрудничать со специалистами различных специальностей: дерматологом, онкологом, психотерапевтом, семейным врачом, эндокринологом. Несмотря на полученный положительный клинический эффект, необходимость динамического наблюдения за течением заболевания обусловлена возможностью рецидивов эрозивно-язвенной формы красного плоского лишая и его злокачественной трансформации.

Ключевые слова:
синдром Гриншпана–Потекаева, полость рта, признаки и симптомы.

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In the clinical practice, the doctors often have to diagnose the erosive-ulcerous form of lichen planus of the buccal mucous membrane, hypertensive disease and diabetes mellitus. The combination of these diseases in one patient is a rather unique pathology and it is called Grinspan-Potekaev syndrome [1]. The epidemiology of the disease is poorly studied [1]. The results of the study conducted by Yu. V. Molochkova et al. indicate that among 338 patients with lichen planus, only one patient (1.23 %) was diagnosed with Grinspan–Potekaev syndrome [2]. The components of the symptom group are closely connected. Excessive blood pressure on the vessels walls of the pancreas contributes to the development of diabetes mellitus that affects further cortical-diencephalic interaction, which is at the core of hypertensive disease and is the initiating agent in the development of lichen planus [3].

Grinspan–Potekaev syndrome is a rare pathology facing dentists in practical activity. That is why the detailed analysis of the disease clinical course peculiarities is important and urgently needed.

Aim

To analyze the clinical course of Grinspan–Potekaev syndrome on the buccal mucous membrane using the example of a patient treatment in the dental clinic Denta Plus (Zhytomyr city).

Materials and methods

Bibliosemantic method (analysis, correlation, comparison, generalization, systematization of scientific literature on the studied issues), system analysis and logical gene-

ralization (for developing the algorithm of diagnostics and treatment), medical documentation analysis of the patient, who sought medical assistance in the dental clinic Denta Plus, were used.

Results

A 59-year-old woman sought medical assistance in the dental clinic Denta Plus. The examination began from the subjective signs of the disease evaluation. Details regarding the duration of the disease and the periods of remission, the relapse rate, the presence of initiating agents and the previous treatment efficacy were clarified by medical history taking. Subjective sensations in the oral cavity and changes related to the general well-being were considered. Previous and comorbid diseases were also determined.

At the first visit, the patient complained of constant pain in the oral cavity worsened by eating, speaking and swallowing, and epigastric burning. Among the general symptoms were weakness, fatigue, anxiety, annoyance, sleep disturbance.

The patient reported the disease duration over the past number of years with periodic exacerbations during the visit. There was no clear seasonal occurrence. By the moment of presentation, the remission period duration had reduced and the relapse rate had increased. At the disease onset, the patient complained of only dryness, burning sensation and discomfort in the buccal mucosa followed by painful nonhealing erosions and ulcers, causing difficulties in eating, brushing teeth, communicating with the people around. The patient associated the disease progress with stressful situations and psychological factors (domestic problems and work issues). Her medical history revealed

among the previous and comorbid diseases that she was having hypertensive disease and gastrointestinal diseases. There was no history of any harmful habits or allergic events.

Pain intensity was assessed using a visual analog scale and registering complaints before, in the process and after the treatment. The scale is a continuous horizontal line ranging from “no pain” (on the left) to “most severe pain” (on the right) [4]. The patient was requested to mark a point on the line corresponding to the perceived pain intensity on examination. Measuring the distance between the marks “no pain” and the mark pointed by the patient, the numerical subjective changes in the oral cavity were scaled.

The scheme of examination consisted of a comprehensive clinical examination with a complex of additional methods, including consultations of interdisciplinary specialists (dermatologist, oncologist, family therapist, psychotherapist). In the process of case follow-up over the disease course, it was recommended to test blood glucose level and a consultation with an endocrinologist. With the aim of the diagnosis verification, test for detecting antibodies against hepatitis B, C viruses, HIV, serological tests, general and biochemical blood analyses as well as clinical urine analysis were performed. The material from the oral cavity was examined for the fungi presence of *Candida* species.

During the clinical examination, special attention was paid to the general appearance; the skin cover was carefully examined, focusing on the flexor surface of the forearms and the skin of the lower legs. A consultation with a dermatologist confirmed and convinced that the lesions were observed only on the buccal mucous membrane. No pathological changes were revealed on the skin, raising difficulties at the initial stage of making a diagnosis.

The oral cavity was examined using an established procedure while applying the standard kit of dental instruments. During examination, the attention was paid to the oral mucosal color and moisture; the presence of pathological elements was detected. The lesions were evaluated in sufficient detail: location, number, shape, size, boundaries and perifocal inflammation processes were determined. The pathological elements were detected simultaneously in some areas of the oral cavity: on the buccal mucosa symmetrically along the tricuspid occlusion line, on the lateral side of the tongue on the left and in the retromolar area on the left. These were multiple ulcers and erosions of various shape, depth and size on the sore and oedematous buccal mucous membrane. Erosions and ulcers were covered with fibrinous pellicle and bled easily upon touch. The area of erosive and ulcerative elements varied. On the right buccal mucosa, the pathological focus was of 5.8 mm² on average, on the left buccal mucosa, it was of 6.4 mm², on the left lateral surface of the tongue, it amounted to 7.1 mm², and in the left retromolar space, it was of 2.3 mm². The area of inflammation and the area of erosions and ulcers were determined using the formula of an irregular circle

$$S = (\pi \times d_1 \times d_2)/4 \text{ in cm}^2,$$

where d_1 is the smallest diameter; d_2 is the largest diameter, π is a constant equal to 3.14. The attention was drawn not only to the affected areas, but also to the condition of the oral floor, edges, tissue density, and regional lymph nodes were palpated.

For ruling out the processes associated with malignant changes, a consultation with an oncologist was followed by morphological examinations providing the conclusion regarding the findings, which stated that the signs of malignant growth were not detected.

Papules of whitish-grey color were noticed among the lesions, confluent papules caused ambiguous picture (“fern pattern”). Such equivocal findings (within the exacerbation period) also complicated the process of making the diagnosis.

On dental examination, the oral cavity was not sanitized, a large amount of soft plaque and plaque deposits were identified, which was the basis to determine hygienic indices (Fyodorov and Volodkina, Green and Vermillion). Before the treatment, the Fyodorov and Volodkina index amounted to 2.9 points (poor level of oral hygiene), the Green and Vermillion index was 1.8 points (unsatisfactory level of oral hygiene). The Schiller–Pisarev’s test was positive and indicated the presence of inflammatory reaction.

The patient was examined by a psychotherapist due to the presence of corresponding complaints. In developing a medical and preventive plan of interventions, the prescriptions and recommendations of the specialist were taken into account.

The patient had no complaints specific to diabetes mellitus, so only in the process of case follow-up and treatment, blood glucose and glucose profile were measured, and the patient was referred to an endocrinologist who diagnosed type 2 diabetes mellitus.

A family physician examined the patient and assigned a therapy for hypertensive disease and gastrointestinal diseases.

The antibodies to hepatitis B, C viruses, HIV were not detected, the serological reactions were negative, clinical urine and blood analyses, blood biochemical values were within the physiological range.

A complete physical examination allowed diagnosing Grinspan–Potekaevev syndrome (erosive ulcerous form of lichen planus with focal damage of the buccal mucous membrane (L.43.8), hypertensive disease (I10), diabetes mellitus (E.11).

Factors underlying the exacerbations, the lesion size and location, the overall health status of the patient, the presence of periodontal pocket infections and previous treatment were considered in developing the plan of therapeutic and prophylactic interventions.

The erosive-ulcerous form of lichen planus of buccal mucous membrane was managed in connection with the concomitant pathology by the family physician, endocrinologist and psychotherapist.

The drug therapy was relatively divided into systemic and topical treatments. The following drugs were administered: “Gidazepam” 0.02 g three times a day (sublingually), “Loratadine” 10 mg once a day, “Methyluracil” 0.5 g three times a day, vitamin-mineral complex “AlfaVit Classic” 1 tablet of each type (No. 1, No. 2, No. 3) once a day with food (an interval between drug intake was 4–6 hours).

Particular attention was given to the local therapy, which included: mouth rinsing with lysozyme, solution “Citalum” 15 drops in a glass of water, oral baths with chamomile and sage decoctions, applications of “Prednisolone”, “Solcoseryl” creams, soaking therapy with wild rose oil and sea

buckthorn seed oil. Anesthesin dissolved in sunflower oil was used for pain relief. 3–6 times a day, the patient treated herself the affected areas with the indicated drugs applying a thin coat on the affected areas at regular intervals. The treatment course lasted 4 weeks.

At the first visit, tooth cusps were flatted, soft plaque was removed and the patient was trained on the rules of oral care. Within the period of exacerbation, a protective diet was recommended (taking to account somatic pathology), ultrasoft toothbrushing, hygienic tooth paste, rinse. Full oral rebridgement including prosthodontic treatment was performed in the remission period.

As a secondary prevention, it was recommended to regulate the schedule of work and rest, to avoid psycho-emotional overload and to follow the oral hygiene rules carefully. In the course of treatment, the patient was continuously interviewed in order to develop motivation to constant preventive examinations (even during the remission period).

The clinical treatment efficacy was evaluated by means of assessing complaints and lesions on the buccal mucous membrane. The control was carried out within one month and the examinations were further conducted weekly.

The complex therapy aimed at regeneration of lesions on the buccal mucous membrane, recovery of arterial pressure and blood glucose level helped to improve the general well-being and to transform the course of erosive-ulcerous form of lichen planus to exudative-hyperaemic one.

Discussion

Nowadays, the issues related to aetiology, pathogenesis, clinical manifestations of lichen planus, modern diagnostic methods and approaches to systemic and topical therapy are sufficiently well covered in scientific literature [1,5,6]. However, among the aspects that need further study, it should be noted the establishment of clinical interrelations of lichen planus with comorbid psychosomatic disorders [7].

Grinspan-Potekaev syndrome is a rare disease facing dentists. The disease is characterized by trilogy, the constituent parts of which are: hypertensive disease, diabetes mellitus, and erosive-ulcerous form of lichen planus [1]. Potekaev N. N. et al., Numan Kekten et al., M. O. Ishkov et al. described isolated clinical cases of a symptom group [8–10]. The typical lesions of the skin, buccal mucous membrane in combination with hypertensive disease and diabetes mellitus were indicated in the described cases.

In the clinical case examined by us, the typical areas of skin lesions were absent and the “fern pattern” on the buccal mucous membrane at the beginning of treatment was unclear presenting difficulties in the process of differential diagnostics conducted between leukokeratosis, exanthematous lupus erythematosus, allergic lesions, erosive lesions in cases of vesicular fever, recurrent aphthous stomatitis. The antibody test to hepatitis B, C viruses, HIV was conducted with the aim of diagnosis verification.

Eva Maria Otero-Rey et al. indicate that lichen planus of the oral cavity is a potentially malignant disorder with a capacity for malignant transformation [11]. One form can quickly transform into the other, which is more severe and dangerous for patient's health or vice versa (in case of timely

diagnosis and appropriate treatment). Erosive-ulcerous form of lichen planus of the oral cavity is especially threatening due to anatomic-physiological peculiarities. The long-lasting presence of nonhealing erosions and ulcers in the oral cavity can lead to the development of malignant tumors. That is why during initial examination of the patient the oncologist consultation was recommended. Regardless of the fact that there were no signs of malignant growth on the oncologist conclusion, the cancer alertness accompanied the entire treatment process. The attention was constantly focused on the area and depth of the lesions, the density of the surrounding tissues and the bed of the eroded surface were evaluated.

The affected areas in case of lichen planus (in the examined clinical case) were located on the buccal mucosa (on the symmetrical sides) along the dental occlusion line, lateral surface of the tongue on the left, in the retromolar area on the left being steadily injured by poor-quality fillings, edges of broken teeth, calcified debris. Non-sanitized oral cavity is not the ultimate cause of lichen planus, but the irritative agents complicate the pathologic processes and slow down the regenerative processes in damaged tissues. For the patient examined in the dental clinic, a complete elimination of local traumatic factors during the exacerbation period was not possible. However, the issues related to the oral sanitation were gradually solved, taking into account the processes of erosion and ulcer epithelialization, pain senses, general well-being. During the first visit, the soft plaque was removed, and the patient was trained on the rules of oral care. Personal hygiene products were chosen and the recommendations regarding the diet were given. Particular attention was paid to orthopedic methods and choice of materials at the final stage of the treatment.

Nowadays, there is no common hypothesis regarding the etiology and development mechanisms of lichen planus [6]. Namely, this causes difficulties within the treatment process. The scientists associate the onset of affected areas with the genetic predisposition or abnormalities, autointoxication processes. Besides there are viral, infectious, neuroendocrine, toxicoallergic, immune-allergic and other theories that partly disclose pathogenetic links of the disease [5]. Lichen planus is pluricausal disease that is why it is necessary to find out the possible causes of the disease development in certain patient for their elimination or reducing the effect. The related specialists help in overcoming this challenge.

In the clinical case presented, the weight was given to the fact that the patient associated exacerbation periods with domestic problems and work issues. At the same time, with the affected areas onset in the oral cavity, the general well-being deteriorated and the general diseases worsened. Namely, these data indicated the need of family therapist and psychotherapist consultations. Delayed regeneration processes at the initial stage necessitated to test blood glucose level. In the clinical case described by Yu. V. Molochkova et al., diabetes mellitus was also diagnosed within the treatment period [3].

Thus, the conducted complex of diagnostic measures allowed to determine the diagnosis (Grinspan–Potekaev syndrome) and to develop and apply complex medicamentous therapy, which positively influenced the general

well-being, promoted the transformation of erosive-ulcerous form of lichen planus into less aggressive and safer exudative-hyperemic one.

Conclusions

1. The described case of Grinspan–Potekaev syndrome allowed to analyze the clinical manifestations of the disease in the oral cavity within the exacerbation phase and to examine the components of diagnostic and treatment process.

2. The treatment response in Grinspan–Potekaev syndrome (with isolated manifestations of lichen planus on the buccal mucous membrane) depends not only on a dentist competence but also on cooperation between other specialists: dermatologist, oncologist, psychotherapist, family physician, and endocrinologist.

3. Despite the obtained positive clinical effect, the need to follow-up the further course of the disease is associated with the possibility of erosive-ulcerous form of lichen planus relapses and its malignant transformation.

Prospects of further studies. The exchange of clinical experience on treatment and diagnostics of rare pathology, namely Grinspan–Potekaev syndrome, will allow to establish the most optimal paths to slow the disease progression.

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