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ORAL MANIFESTATIONS IN DISCOID AND SYSTEMIC LUPUS ERYTHEMATOSUS

I. CLINICAL INVESTIGATION

by

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The oral manifestations of lupus erythematosus (i.e.) were first described in 1861 by *Bazin*, but in 1901 a more detailed description of the oral lesions was given by *Capelle*. Later on, *Kren* 1907, *Haslund* 1916, *Folpmers* 1937, and *Ebrard* 1955 contributed additional descriptions of oral lesions. Apart from this, few reports could be found in the literature concerning the morphology and natural course of oral lupus erythematosus.

Three types of lupus erythematosus are generally recognized. The most frequent type is the chronic discoid lupus erythematosus with affections especially on the skin of the nose and the malar eminences (butterfly pattern). The scalp, ears, and hands may be involved too. The cutaneous lesions consist of well-defined erythematous patches, adherent keratotic scaling, and follicular plugging. In addition, older lesions show atrophic scarring.

The subacute type often shows lesions in the face and lesions located to thorax and extremities as well. They appear as erythematous, coalescing areas with moderate scaling. Systemic

symptoms such as malaise, fever, and leucopenia are often present.

The acute systemic type is dominated by severe systemic symptoms of fever and pain in muscles and joints as well as blood changes, such as leucopenia and hypergammaglobulinemia. The i.e. phenomenon will usually be positive. The cutaneous eruptions often begin in the face as a diffuse erythema and edema which, later on, spreads widely over the body. Sometimes, skin lesions do not appear at all. The disease often ends lethally.

Table I
Frequency of oral lesions in lupus erythematosus

Author	Year	Type of lupus erythematosus	Number of patients with cutaneous lupus erythematosus	Number of patients with lesions on oral mucous membrane and vermillion border	
				Number	Percentage
Smith	1906	discoid	46	11	24
	1906	systemic	10	4	40
Culver	1915	discoid	38	6	16
Haslund	1916	discoid?	162	23	14
Monash	1931	discoid?	22	11	50
Winsiffer	1934	discoid?	103	18	17
Poehlman	1936	discoid?	37	2	5
Shearn & Pirofsky	1952	systemic	34	13	38
Soffer & Bader	1952	systemic	18	9	50
Dubois	1953	systemic	62	7	11
Jessar et al.	1953	systemic	44		18
Orban & Wentz	1960	discoid?	104	19	18
Marten & Blackburn	1961	discoid	51	3	6
Sørensen	1962	systemic	11	4	36

Concerning the frequency of oral lesions in i.e., reports in the literature vary, Table I. (In five reports the type of i.e. was not mentioned, but from the clinical descriptions they appear to be of the discoid variety). *Ebrard* 1955 reports that the oral lesions develop most frequently after the skin eruptions, while *Traut-*

mann 1911 generally finds that the lesions occur at the same time.

A number of authors have reported cases of oral i.e. located to the vermillion border of the lower lip without cutaneous lesions (*Ebrard* 1955, *Haslund* 1916, *Kren* 1907).

Table II

Development of cancer in oral discoid lupus erythematosus. Data from 16 cases reported in the literature

Author	Year	Sex	Age in Years	Duration of oral discoid lupus erythematosus (in years)	Location of oral cancer	Type of cancer	Previous x-ray treatment
Taylor	1898	♂			Lower lip		
Kreibich	1900	♂	36	2½ years	Upper lip	Squamous cell carcinoma	
Baumm	1907	♂	44		Upper lip	Carcinoma	
Pautrier & Fage	1909	♂	47	11 years	Lower lip	Squamous cell carcinoma	
Schwartz	1918	♂		15 years	lip	Epithelioma	
Nobl & Lövenfeld	1926	♂	56	6 years	Lower lip	Squamous cell carcinoma	—
Arzt	1926	♂	46		Lower lip	Squamous cell carcinoma	—
Geserowa	1929				Upper lip	Carcinoma	
Riehl	1930		57		Upper lip	Squamous cell carcinoma	
Ullmo	1932	♂	36	13 years	Lower lip	Squamous cell carcinoma	—
Tischnenko	1932	♂	21		Lower lip	Squamous cell carcinoma	—
Dörffel	1936	♀	69		Lower lip	Squamous cell carcinoma	
Folpmers	1937	♂	60		Lower lip	Squamous cell carcinoma	—
Périn et al.	1940	♂	28	2 years	Lower lip	Squamous cell carcinoma	—
Berggren	1941			3 years	Lower lip		+
Andreev et al.	1961	♂	54	21 years	Upper lip	Squamous cell carcinoma	—

Discoid lupus erythematosus*Vermilion border*

The vermilion border of the lower lip is most frequently involved (*Haslund 1916, Pautrier 1936*). The lesions often spread to the lips from the skin but may develop as a primary lesion. The first stage is characterized by a diffuse or localized erythema with a few telangiectases. After some time an adherent, indurated keratotic scaling appears (*Ebrard 1955, Folpmers 1937*). The fully developed lesion is described as an atrophic lesion surrounded by a keratotic border (*Haslund 1916, Kren 1907*). Apparently, the typical plaque develops from a confluence of smaller lesions. This plaque may involve either a small part of or the entire vermilion border and even extend to the cutaneous part of the lip. The vermilion border generally shows edema and slight eversion.

Some authors have described carcinomatous changes in discoid lupus erythematosus located in the vermilion border, Table II. A number of reported cases are not included in Table II, because the diagnosis of cancer was not verified histologically (*Beurmann & Laroche 1909, Graham-Little 1932, Jarke 1922, Oliver 1930*).

Oral mucosa

The oral lesion begins as an erythematous, sometimes slightly elevated infiltrate without induration. Demarcation of the lesion is not quite sharp. Unlike the lip lesions, those of the mucosa of the oral cavity have no stage of scale formation. Keratinization of the mucous membrane develops and is usually manifested as an atrophic red lesion surrounded by a keratinized narrow zone (*Haslund 1916, Kren 1907*).

The central area may turn into a superficial, painful ulceration. The lesions of the oral mucosa usually appear bilaterally and are frequently located opposite the molars.

Tongue lesions are associated with an atrophy of the papillae (*Ebrard 1955*). Additionally, an intense fissuring of the tongue may be seen.

In palatal lesions the hard palate is most frequently involved. Cases of discoid l.e. affecting the gingivae have been reported (*Archard et al. 1963, Cooke 1953, Haslund 1916, Lite 1953*).

Subacute lupus erythematosus

Oral lesions in this form of lupus erythematosus have, as far as the present author is aware of, not been described in the literature.

Systemic lupus erythematosus

Schuermann (1958) maintains that the clinical picture changes more rapidly in systemic l.e. than in discoid l.e. and furthermore that in the acute, systemic type the hyperemia, edema, and extension are more pronounced. There is a greater tendency to ulceration and bleeding. Petechiae and superficial ulcerations, surrounded by a red halo, are often present (*Dubois* 1953). Sometimes the vermilion border is completely covered by haemorrhagic crusts (*Schuermann* 1958). Oral lesions appear especially in the acute phase of the disease. *Rauch* (1959) collected 8 cases from the literature with systemic l.e., which, at the same time, demonstrated bilateral swelling of the parotid glands. Four of them had xerostomia. Superimposed oral moniliasis due probably to low resistance of the patients or prolonged steroid therapy may complicate the picture. Regarding treatment *Soffer & Bader* (1952) found satisfactory effect of general therapy with steroids.

OWN INVESTIGATIONS

Sixteen patients with oral lupus erythematosus were examined. All of them had skin lesions. 9 cases were of the discoid type, 3 cases of the subacute form, and 4 cases of the acute, systemic type.

Sex and age of the patients and location of the oral lesions are tabulated in Table III.

In the discoid l.e. the early lip lesions located to the vermilion border showed keratotic scaling, Fig. 1. The fully developed lesions showed a polycyclic demarcation consisting of a small keratinized border-zone composed of small, radiating white striae. The central part of the lesions showed small, white dots and lines intermingled with red atrophic areas, Fig. 2. The lines were arranged in a lace-like or parallel pattern. The central area frequently changed into a crusted ulcer or fissure, which often bled.

Table III
Patients with oral lupus erythematosus

Patient	Sex	Age in years	Type of lupus erythematosus	Localization of oral l.e.	Biopsy
1. E. J.	♀	37	Discoid	Buccal mucosa bilaterally	+
2. M. M.	♀	40	Discoid	Vermilion border of upper lip	—
3. I. N.	♀	34	Discoid	Vermilion border of lower lip	+
4. E. S.	♀	34	Discoid	Buccal mucosa bilaterally	+
5. M. M.	♀	28	Discoid	Inner surface of lower lip	+
6. T. N.	♀	27	Discoid	Gingiva	+
7. E. B.	♀	35	Discoid	Vermilion border of lower lip, buccal mucosa	+
8. E. J.	♀	46	Discoid	Vermilion border of upper lip	—
9. B. J.	♀	44	Discoid	Buccal mucosa	+
10. E. F.	♀	44	Subacute	Vermilion border of upper and lower lip, buccal mucosa bilaterally, hard palate, tongue	+
11. G. W.	♀	58	Subacute	Buccal mucosa bilaterally	+
12. P. B.	♀	38	Subacute	Vermilion border of lower lip, buccal mucosa bilaterally	+
13. A. R.	♀	34	Systemic	Buccal mucosa	+
14. K. N.	♀	46	Systemic	Vermilion border of lower lip, buccal mucosa bilaterally, hard and soft palate	+
15. E. R.	♀	26	Systemic	Vermilion border of lower lip, buccal mucosa bilaterally	—
16. E. P.	♀	37	Systemic	Buccal mucosa, hard palate	+

In the oral mucosa a very early lesion affecting the buccal mucosa was discovered, Fig. 3. In spite of its small size this lesion had already developed a small central atrophic area, surrounded by a keratotic border. The pronounced hyperemic zone around the lesion possibly indicates a state of active spreading.

The typical lesion in the oral cavity showed a central depressed red atrophic area with small white dots and lines. This area was surrounded by a 2 to 4 mm wide elevated white keratotic zone, peripherally dissolving into small, parallel white lines, Fig. 4. Outside this zone some hyperemic radiating vessels could be recognized. The lesions presumably extend by peripheral growth and when several of them coalesce, a polycyclic figure appears, Fig. 5.

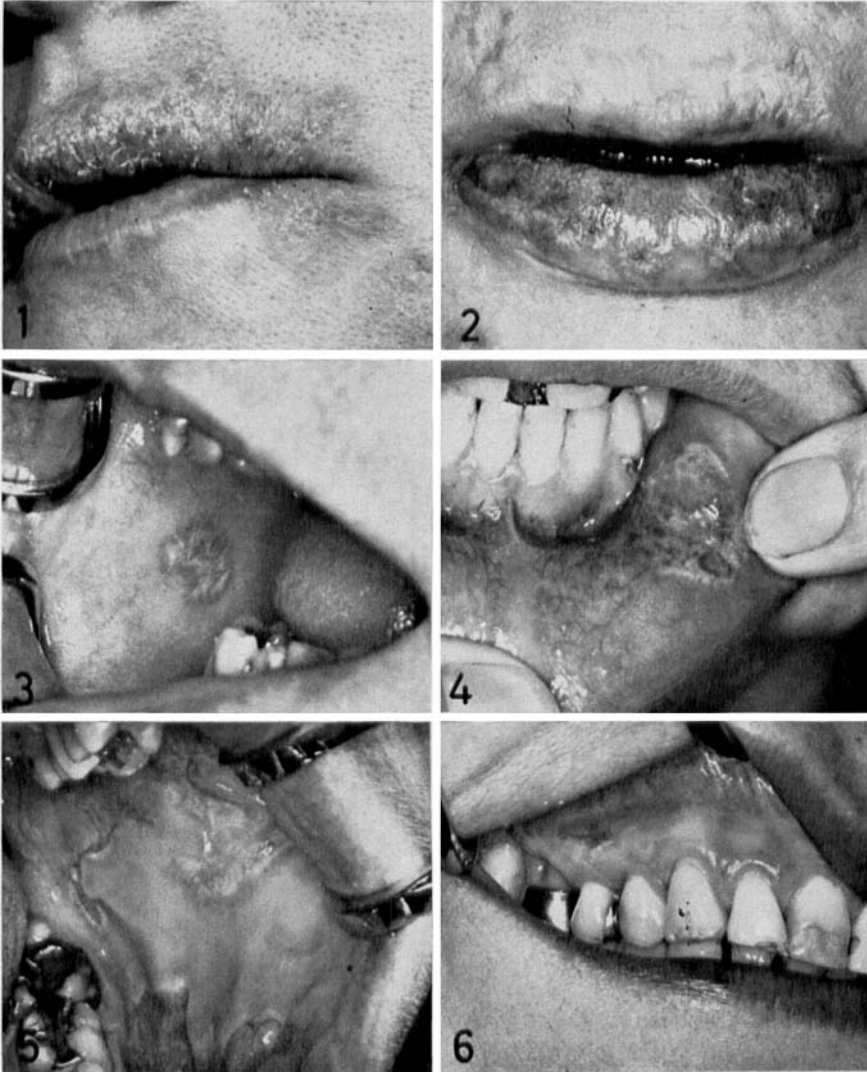


Fig. 1. Discoid l.e. lesion of vermillion border and adjacent skin showing erythema and keratotic scaling.

Fig. 2. Fully developed discoid lesion of lower lip. Note the marginal zone and the central atrophic area with keratotic spots and lines.

Fig. 3. Early discoid lesion of buccal mucosa. Note the atrophic central area, the keratotic marginal zone, and the red halo surrounding the lesion.

Fig. 4. Typical discoid l.e. lesion. Note the characteristic marginal zone composed of small white lines and the central atrophic area.

Fig. 5. Polycyclic discoid lesion located to buccal mucosa formed by coalescence of several lesions.

Fig. 6. Discoid l.e. located to alveolar gingiva.

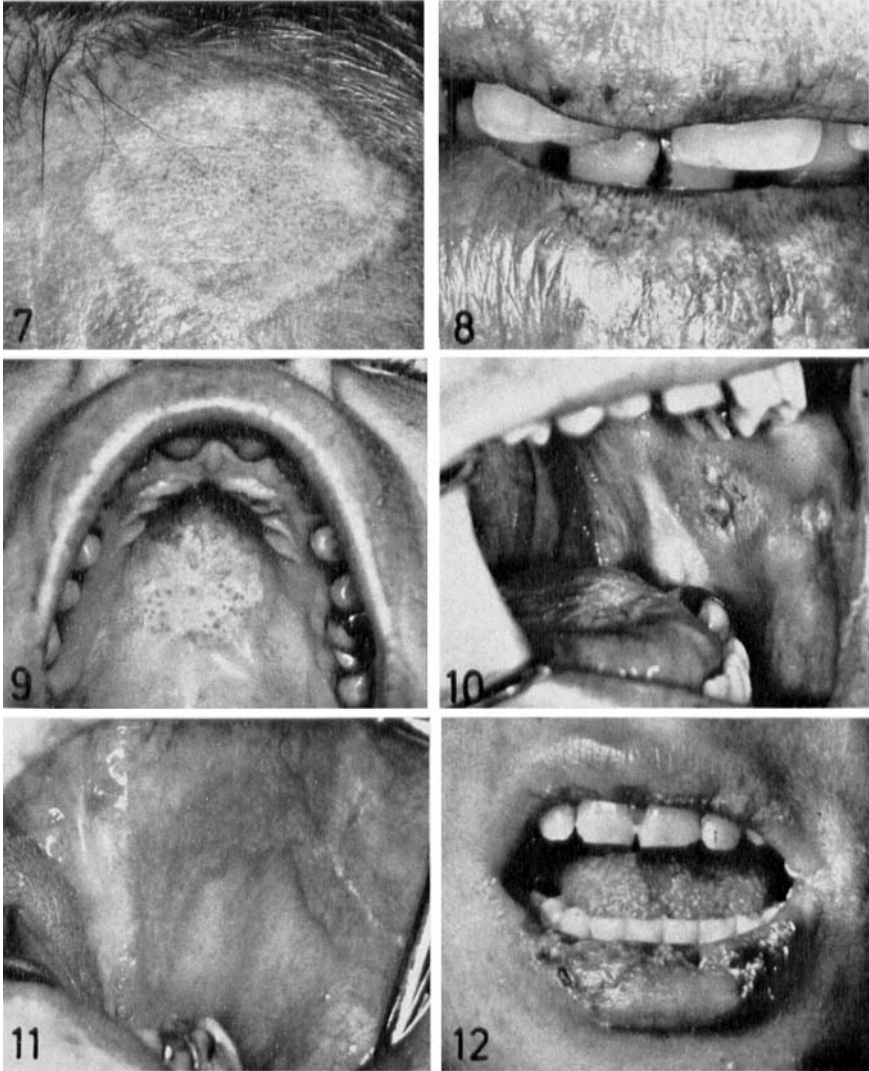


Fig. 7. Discoid lesion located to the forehead. Note the border zone and the central atrophic area.

Fig. 8. Subacute l.e. located to lower lip.

Fig. 9. Subacute l.e. affection located to hard palate. Note the similarity to leucoplakias.

Fig. 10. Subacute l.e. of buccal mucosa in the same patient as Fig. 9. Note the central ulceration.

Fig. 11. Systemic l.e. lesion of buccal mucosa consisting of inspecific erythema.

Fig. 12. Lip lesion in systemic l.e. showing ulceration and hemorrhagic crusts.

A gingival lesion with a similar morphology was found, Fig. 6. The buccal mucosa and the vermilion border were the areas most often affected, Table III.

Examination of cutaneous lesions revealed the same characteristic border-zone and central atrophy as found in the oral lesions, Fig. 7. As a result of treatment with anti-malarial drugs there was a complete disappearance of the oral discoid lesions in 2 cases and a partial improvement in two others. In the remaining cases there was no benefit from general treatment with anti-malarial drugs in spite of good effect on the skin lesions. As a result of treatment with chloroquine® one patient developed a very extensive pigmentation of the vermilion border and oral mucous membrane (*Zachariae 1963*).

Two patients complained of slight pain in their oral lesions with aggravation during the menstrual period.

Most of the patients were followed over a period from 1 to 5 years and in none of these did the lesions show malignant degeneration.

In the subacute affections there was a greater extension of lesions in the oral cavity and greater tendency to ulceration.

One case involved the vermilion border with central atrophy surrounded by a keratotic zone, Fig. 8, but usually the lesions were irregularly demarcated keratotic areas often showing ulcerations, Figs. 9 and 10. In one case a marked fissuring of the tongue was found.

Cases of systemic i.e. showed erythematous areas with superficial ulcerations but no keratinization of the oral mucosa, Fig. 11.

In one patient a considerable crusting of the lower lip was found, Fig. 12.

One case reported below showed oral lesions as the first sign of the disease.

Case report

Case no. 16: A 37-year-old woman was admitted to the Dental Clinic on July 4th 1961. She had had an ulceration in the hard palate for one month. During this time she had been treated for anemia. She had no skin lesions. Clinical examination revealed

an atrophic area with a keratotic border. After one year the oral lesions were unchanged, but at that time skin lesions appeared on the nose and the cutaneous part of the upper lip. The patient was admitted to the dermatologic department. The skin lesions were suspected to be those of lupus erythematosus and were treated with periactin®. Two months later the patient was hospitalized with anemia, albuminuria, hematuria, and edema.

At that time laboratory values were as follows: Hbg. 48 %. E.S.R. 80 mm. Gammaglobulin 1.48 %. The patient's condition deteriorated and death occurred after two weeks.

Autopsy diagnosis of systemic l.e. was based on typical renal changes.

DISCUSSION

In the present material it has not been possible to determine the frequency of oral lesions in l.e. patients, but previous investigations state that about 20 % of patients with the discoid type show oral lesions, Table I. Oral lesions appear to be slightly more frequent in the systemic type than in the discoid type, Table 1. The distribution of sex and age in this material is the same as that found by previous investigators (*Culver* 1915, *Smith* 1906, *Sugar* 1954). In the present study the buccal mucosa and the vermilion border were the predilection sites. The same was found by *Monash* (1931) and *Sugar* (1954).

Similar morphology of the oral lesions in the discoid l.e. as described in this investigation has been reported by *Haslund* (1916) and *Kren* (1907). In the present material oral lesions of the subacute and systemic types show greater extension and more frequent changes of the clinical picture than does the discoid type. This difference is well known as concerns the cutaneous lesions too. The oral lesions appear to give few symptoms, *Haslund* (1916) and *Kren* (1907).

In two cases symptoms in discoid oral lesions were related to the menstrual cycle. A similar correlation regarding cutaneous lesions was described by *Pilsbury et al.* (1960).

The discoid oral lesions are reported to have a marked resistance to general treatment (*Pierini* 1956). The present study confirms this except for two cases.

The intimate relationship between oral and cutaneous discoid lesions was proved by the great similarity of the initial and fully developed lesions and their way of progression. Both oral and cutaneous lesions showed the characteristic keratotic border-zone which encircles an atrophic area. Scaling found on the vermilion border was absent on the mucous membrane probably because of its special histologic structure, and the moistening from saliva of this area.

The rate of development of cancer in cutaneous discoid i.e. varies from 0.5 % to 3.6 % (*Lehmann 1936, Luschtschizkij 1936, Schwarz 1953, Wander 1921*). No reports have been published on the frequency of malignant change in oral lesions. All cases of cancer in oral discoid lesions were located to the vermilion border, especially to the lower lip, Table II. There has been no report on malignancy in the oral mucous membrane lesions. Males are more prone than females to develop cancer in oral lesions. 12 male and one female cases have been reported. This does not agree with the ordinary sex distribution in discoid i.e. where females are most often affected (*Herzberg 1950*). A possible explanation is that males show a greater tendency to develop lip cancer than females (*Cross et al. 1948*). Previous radiation treatment cannot be blamed as a causative factor in malignant changes in the oral discoid lesions, as it was used in only one case, Table II.

Andreev et al. (1961) propose that just like in xeroderma pigmentosum, the demonstrated photosensitivity of discoid i.e. patients might predispose them to malignant degeneration.

SUMMARY

A review of the literature concerning oral lesions of lupus erythematosus is presented, and a material of 16 cases with oral lesions is reported. 9 cases were of the discoid type, 3 cases of the subacute, and 4 cases the systemic type.

Skin lesions were present in all 16 patients. The buccal mucosa and the vermilion border were most often involved. The discoid type showed characteristic oral lesions consisting of central red atrophic areas surrounded by white keratotic borderlines. The similarity in morphology of oral and cutaneous discoid lesions

is pointed out. The oral lesions of discoid and subacute l.e. have a very chronic course and often resist systemic treatment.

One case of systemic l.e. with oral lesions as the first sign of the disease is reported.

RÉSUMÉ

MANIFESTATIONS BUCCALES DANS LE LUPUS ÉRYTHÉMATEUX DISCOÏDE ET DANS LE LUPUS ÉRYTHÉMATEUX DISSÉMINÉ I. ÉTUDE CLINIQUE

L'auteur présente une revue de la littérature concernant les lésions buccales du lupus érythémateux et rend compte d'un ensemble composé de 16 cas comportant des lésions buccales. 9 des cas étaient du type discoïde, 3 du type subaigu, et 4 du type disséminé.

Les 16 patients présentaient tous des lésions cutanées. La muqueuse des joues et le bourrelet rouge de la lèvre étaient le plus souvent atteints. Le type discoïde présentait des lésions buccales caractéristiques consistant en zones centrales atrophiques rouges entourées d'une ligne de démarcation kératosique blanche. L'auteur souligne la similitude de la morphologie des lésions discoïdes buccales et cutanées. Les lésions buccales du l.e. discoïde et du l.e. subaigu ont une évolution très chronique et résistent souvent au traitement général.

L'auteur rapporte un cas de l.e. disséminé dont le premier symptôme avait été une lésion buccale.

ZUSAMMENFASSUNG

ORALE MANIFESTATIONEN DER LUPUS ERYTHEMATODES DISCOIDES UND DISSEMINATUS

I. KLINISCHE UNTERSUCHUNGEN

Die Literatur betreffs oraler Manifestationen des Lupus erythematodes wird besprochen, und ein Bericht über Untersuchungen des Verfassers von 16 Fällen mit oralen Affektionen wird gegeben. 9 der Fälle waren Lupus erythematodes discoides, 3 Lupus erythematodes subacutus und 4 Lupus erythematodes disseminatus.

Affektionen der Haut waren an allen Patienten zu finden. Die Mundschleimhaut und der Lippenrot waren am häufigsten und am meisten angegriffen. Der Lupus erythematodes discoides ma-

nifestierte sich in typischen Fällen als zentrale, rote, atrophische Gebiete mit einer weissen, keratotischen Randzone.

Beim Lupus erythematodes gibt es dieselbe Morphologie für Affektionen der Mundschleimhaut und der Haut. Die orale Affektionen des Lupus erythematodes discoides und subacutus haben einen sehr chronischen Verlauf und widerstehen oftmals innere Behandlung.

Ein Fall von Lupus erythematodes disseminatus mit oralen Affektionen als erstes Zeichen der Krankheit wird besprochen.

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