SECONDARY LYMPHEDEMA OF LIPS AS A SYMPTOM OF OROFACIAL LESIONS

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The study involved 18 patients with the secondary lymphedema of lips, Melkersson-Rosenthal syndrome, Granulomatous cheilitis of Miescher, Crohn’s disease, and chronic odontogenic periapical inflammation. Based on the data of clinical and ultrasound examinations authors have established the similarity of the clinical picture and the course of lymphedema in these diseases. The authors suggest not to consider the secondary lymphedema of lips an independent nosological entity.

Keywords: lips lymphedema, macrocheilitis, Melkersson-Rosenthal syndrome, Granulomatous cheilitis of Miescher, Crohn’s disease.

Lymphedema is a congenital or an acquired disease of the lymphatic system, associated with the abnormality of the outflow of lymph from the lymphatic capillaries and peripheral lymphatic vessels from organs and tissues to the main lymphatic collectors and the thoracic duct, which leads to an increase in the size of the affected organ.

Lymphedema occurs when lymphatic load exceeds the transport capacity of the lymphatic system. The imbalance between the formation of lymph and its outflow occurs in the course of various diseases, including the orofacial pathology. Primary lymphedema occurs more rarely, in idiopathic or acquired vascular malformations, especially in their hypoplasia or aplasia. Secondary Lymphedema usually develops in impairment of lymph transportation due to damage or resection of lymphatic vessels and lymph nodes, infections and radiation [6, 8].

In dental clinics they adhere to the same systematics and consider lymphedema of lips as primary pathology (ICD-10: Q82.0 Hereditary lymphedema. Q18.6 Macrodontia) and secondary pathology (ICD-10: I89.0 Lymphedema, not elsewhere classified) [14].

The literature describes secondary lymphedema of lips in a number of diseases, one of the leading clinical symptoms of which is macrochilia [4, 7, 9, 12], but there is no information about the features of the clinical picture of swelling of each of these diseases.

The aim of our study was to offer the comparative evaluation of the clinical picture of secondary lymphedema of lips with orofacial lesions.

Materials and methods. Survey of 18 patients aged 51-73 was conducted; among them there were 15 women and 3 men. Patients were sent for consultations to the department with various orofacial pathologies, where the main symptom of disease was macrochilia. Patients underwent clinical, radiological, laboratory and ultrasound examination. Clinical and laboratory tests – according to the generally accepted methods, orthopantomography – on apparatus PDX0771000, ultrasound – on Toshiba «Aplio» of expert class using Doppler ultrasound.

Results and discussion. Analysis of the results of a complete examination of patients allowed determining the Melkersson-Rosenthal syndrome in 3 patients. Among these patients, 2 were directed with the erysipelas face. In 3 patients the syndrome was characterized by the classic symptoms, in 2 – two symptoms. Moreover, one patient formed a triad of symptoms during 25 years. In 2 patients together with the gastroenterologist we diagnosed the «Crohn’s disease». Granulomatous cheilitis of Miescher was diagnosed in 3 patients. The cause of the remaining 8 cases of macrochilia was the local stomatogenic pathology (granulating periodontitis, radicular cyst, periodontal disease with bone pockets, a chronic relapsing labial fissure).

Despite the different genesis of the disease all patients have a common symptom – lips are increasing in size due to their edema. They complained of discomfort in the affected lip, the feeling of heaviness and tightness in it, violation of diction. Pain was absent.

The same type for macrochilia was characteristic for the medical history. As a rule, patients noted cyclic course with sequences of relapse and remission periods. Firstly the edema of the upper or lower lip appeared, which lasted from a few weeks to several months. In remission period the edema did not disappear completely; with each subsequent relapse its strengthening was observed.

During the examination the edema of the upper or lower lip was determined. It was generalized and even or asymmetrical, damaging more than one of the halves of the lips. Edema captured topographically only one or two lips, sometimes spread beyond it. Most often, the skin of the lips had elements of pigmentation due to telangiectasia, slight cyanosis. On the vermilion zone there were signs of trophic disorders – sometimes atrophy and thinning of the skin, peeling, angiectasia (Fig. 1, 2).

Tissues were of tightly-elastic consistency at palpation, painless; after pressing the marks remained. In the lips thickness the small nodules were slightly palpable (small salivary glands).

Ultrasonography has determined the heterogeneity of structure without clear contours, decreased echogenicity, increased vascularization and vasodilatation in lips thickness.

In patients with odontogenic factors as the cause of lymphedema, the signs of apical periodontitis, radicular cysts,
Deep bone periodontal pockets were determined on orthopantomography (Fig. 3).

Data of laboratory examinations were within the age norm. Thus, clinical and ultrasound picture of lymphedema of lips and its clinical course had common features, despite the different genesis of the disease.

The most recognizable in the clinic the Melkersson-Rosenthal syndrome is a rare disease of unknown ethology. Clinical manifestations are characterized by the typical triad of symptoms: swelling of face and lips, peripheral facial paralysis and fissured tongue. According to the literature data, only 75.4% of patients have this syndrome accompanied by the classical triad, the others have one or two symptoms [3, 12]. This is confirmed by our study. Sometimes the syndrome is accompanied not only by the swelling of lips, but also other parts of the face, particularly by isolated edema of the upper eyelid [2].

Among the diseases, having the edema of lips as their symptom, Granulomatous cheilitis of Miescher can also be noted [5, 7]. There is evidence of a combination of granulomatous cheilitis with vulvitis. Moreover, the results of histological studies indicate their similarity [1]. A similar morphological pattern of lips edema in cases of Granulomatous cheilitis of Miescher and Melkersson-Rosenthal syndrome is noted by other researchers [5, 7].

Some authors separately determine the orofacial granulomatosis [10]. It is also a rare disease that is characterized by persistent or recurrent swelling of the soft tissues of the mouth, ulceration, and the presence of the non-caseating granulomas in the tissues thickness. This term was introduced to integrate a range of different disorders, including the Melkersson-Rosenthal syndrome and granulomatous cheilitis (which is sometimes considered a mono-symptomatic form of the Melkersson-Rosenthal syndrome), and is regarded as synonymous of the previously considered disease [11].

**Conclusions.** The similarity of the data of anamnesis, clinical and other methods of research of the secondary lymphedema of lips allow us to consider it symptomatic, and not to allocate this pathology as a separate nosological entity.

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