

OROFACIAL SYMPTOMS RELATED TO BORRELIOSIS – CASE REPORT

Ewa Wolańska-Klimkiewicz¹, Jolanta Szymańska², Teresa Bachanek¹¹Chair and Department of Conservative Dentistry, Medical University of Lublin, Poland²Chair and Department of Paedodontics, Medical University of Lublin, Poland

Wolańska-Klimkiewicz E, Szymańska J, Bachanek T: Orofacial symptoms related to borreliosis – case report. *Ann Agric Environ Med* 2010, **17**, 319–321.

Abstract: Lyme disease (borreliosis) is an infectious disease caused by the *Borrelia* spirochaetes, usually transmitted to humans by the *Ixodes* ticks. It is manifested by a wide spectrum of clinical symptoms varying according to the time elapsed from the infection. The paper describes orofacial symptoms of the disease in a woman who has suffered from a chronic borreliosis for 3 years. Multiorgan clinical manifestations of borreliosis might occur in the masticatory organ and this fact should be taken into consideration in the differential diagnosis of pain by medical and dental practitioners.

Address for correspondence: Chair and Department of Conservative Dentistry, Medical University of Lublin, Karmelicka 7, 20-018 Lublin, Poland. E-mail: zstzach@am.lublin.pl

Key words: borreliosis, human, masticatory organ, pain.

INTRODUCTION

Lyme disease (borreliosis or tick-borne spirochaetosis) is an infectious disease caused by the *Borrelia* spirochaetes, usually transmitted to humans by the *Ixodes* ticks. The genospecies of *Borrellia burgdorferii* that are pathogenic for humans include: *Borrelia burgdorferi* sensu stricto, *Borrelia afzelii* and *Borrelia garinii* that lead to the disease. Borreliosis is a multiorgan disease and, according to the time elapsed from the moment of infection, it is manifested by different clinical symptoms. The genospecies *Borrelia burgdorferi* sensu stricto causes mostly joint borreliosis, *Borrelia afzelii* – chronic atrophic dermatitis and cutaneous lymphoid hyperplasia, and *Borrelia garinii* – neurological symptoms [6].

In the first of the three stages of the disease, the early infection stage, the symptoms are: *erythema migrans* (EM), showing the migration of the spirochaetes in the skin, and the borrelial lymphocytoma (*lymphadenosis benigna cutis*, LBC) in the form of a lumpy, painless, purple-coloured lesion that develops on the ear lobe, nipple or scrotum. Erythema eruptions are accompanied by topical symptoms, such as itching and a burning sensation, as well as pain due to the local neuritis. Migrating erythema is the most common symptom in comparison to other skin lesions

observed in the course of infection [2]. *Erythema migrans* may be caused by all the genospecies of *Borrellia burgdorferii* [9]. In the early localized form of the disease, which is its second stage, further skin lesions (LBC), as well as acute inflammation in the organs such as heart, joints, and central and peripheral nervous system, develop. At this stage, early neuroborreliosis may take the form of meningitis. The inflammation involving the eye choroid coat, liver, testicles and intestines is also possible. The third stage of the disease, i.e. late borreliosis, takes the chronic form developing from one to several years after the moment of infection. This stage involves chronic atrophic dermatitis, chronic arthritis, as well as neurological and psychiatric syndromes; however, damage to the central or peripheral nervous system occurs most frequently [14].

CASE DESCRIPTION

A 50-year-old woman, B. S. reported that while working in the garden, she had been bitten by an insect in the area of the right eye and on the lower leg. An inflammatory response in the form of swelling on the right upper and lower eyelids, cheek and neck developed after 12 hours. Similar, but less pronounced changes occurred also in the lower limb. The woman presented at the emergency

department where she was administered antihistamine drugs. She was recommended the application of compresses with aluminium acetotartrate (Altacet) that, applied topically onto the skin, has an astringent effect relieving pain in the inflammatory focus, softens the epidermis, reduces swelling in tissues, and alleviates the symptoms of inflammation; and an ointment containing mometasone furoate, a mid-strength synthetic corticosteroid with an anti-inflammatory and anti-pruritic effect (Elocom). Two weeks after the application of the described treatment, the local symptoms receded.

Two months after the bite, the general condition of the patient deteriorated. She began to suffer from pain in the joints and muscles, paroxysmal pain in the heart area, weakness and fatigue, conjunctivitis and hypersensitivity to light. Dental symptoms also appeared: paroxysmal bilateral pain in upper and lower teeth. The pain was acute, radiating, and difficult to localise. Clinical and radiological examination of teeth, and of upper and lower maxillary bones, excluded dental causes of pain. Ophthalmological consultation did not determine the cause of chronic conjunctivitis. After 5 months the patient reported pain in the chest in the heart area, radiating towards the left arm, with a concurrent increase in blood pressure (170/105 mm Hg). Cardiologic consultation did not find abnormalities in the electrocardiographic curve. The patient was recommended oral treatment, used in mild forms of cardiac neurosis, with drugs containing aethylium bromisovalerylum, fenobarbitalum, oleum menthae piperite, oleum humuli lupuli, ethanol (Milocardin), magnesium and vitamin B preparation, as well as gastro-resistant tablets containing acidum salicylicum, used in the prevention of ischemic heart diseases (Acard). Despite the treatment the patient continued to suffer from the described complaints. After another 2 weeks, i.e. 5.5 months after the bite, an erythema, ca. 12 cm in diameter, developed on the lower limb. Flu-like symptoms appeared and intensified. Dermatological consultation led to a preliminary diagnosis of the first-stage of borreliosis. Treatment with a macrolide antibiotic was recommended. Despite the fact that antibiotic therapy was repeated twice, the patient began to suffer from neurological complaints: disorders of consciousness, difficulties in naming objects, paralysis of the left side of the face, and numbness in the right cheek.

The serological ELISA test for borreliosis after 7 months after the bite showed a high titre of IgM antibodies: 46.7 BBU/ml, as well as IgG antibodies: 37.3 BBU/ml (a result is considered positive when the antibody titres exceed 11 BBU/ml). A month of therapy with an antibiotic of the doxycycline group (Unidox) and a chemotherapeutic agent of the nitroimidazole derivatives group (Metronidazol) was recommended. During the treatment, other neurological symptoms occurred, such as motor coordination disorders, numbness and formication in the upper and lower limbs, as well as sleeplessness. The patient reported a sensation of dryness and metallic taste in the mouth. Due to the

deterioration of her general condition, the patient was hospitalized. The magnetic resonance examination showed diffuse damage to the CNS.

An ELISA test performed 11 months after the infection showed a low level of both immunoglobulins – less than 5 BBU/ml. As the clinical symptoms persisted, the patient underwent another antibiotic treatment with a drug of the amoxicillin group (Amoksiklav) and the previously used chemotherapeutic agent Metronidazol.

A subsequent ELISA serological test for borreliosis, however, showed an increase in the IgM antibodies titre in the blood serum – 24.577 BBU/ml. The titre of IgG antibodies was 8.142 BBU/ml (the upper limit of the acceptable norm, while the result 9–11 BBU/ml is considered as ambiguous). The results faintly decreased after another hospitalisation and reached: IgG – 5.449 BBU/ml and IgM – 21.014 BBU/ml.

The patient remains under the care of zoonoses, neurological and audiological outpatient clinics because of the persisting complaints characteristic of neuroborreliosis. At present, there are following symptoms related to the masticatory organ: Bell's palsy, the feeling of numbness on one side of the face, acute toothache radiating towards the eye and ear – according to the patient's description: "a boring pain". Those complaints recur periodically and are not caused by any dental factor. When the pain appears, the patient visits the dentist to determine the cause and possible treatment. To date, no dental cause of the paroxysmal pain has been found. For diagnostic purposes, the fillings were removed to apply anti-inflammatory and odontotropic medication. After 2–4 weeks the drugs were replaced with fillings. The response of the dental pulp to vitality tests has always been normal.

DISCUSSION

The described symptoms are typical of neuroborreliosis. However, the course of the disease is atypical. Sometimes the symptoms of the disease pertain to its different stages, or simultaneously affect different organs [2]. In the described case the concurrence of different clinical symptoms might be the cause of diagnostic difficulties. The late diagnosis of the disease was due to the fact that the subsequent symptoms had not been causally connected to those that occurred initially.

The variety of symptoms may be connected to the genospecies coinfection of *Borellia burgdorferii* ticks, as well as to coinfection with other pathogens – viruses, bacteria, protozoans, for which ticks are important vectors. They include tick-borne encephalo-meningitis virus, *Anaplasma phagocytophilum* and *Babesia microti* [7, 8, 10, 12]. A study on the coincidence of 3 pathogens: *Borellia burgdorferii* sensu lato (s.l.), *Anaplasma phagocytophilum* and *Babesia microti* in *Ixodes ricinus* ticks in the territory of the Lublin macroregion shows that coinfection with 2 or 3 pathogens occurred in 2.16% of the ticks. The most

common was the coincidence of *Anaplasma phagocytophilum* with *Babesia microtii*; a similar result was obtained for the coincidence of *Borrelia burgdorferi* s.l. with *Anaplasma phagocytophilum* [15]. It is believed that coinfections with bacteria or protozoans in a patient with borreliosis may either aggravate the course of the disease [1, 13] or lead to its milder form [8]. This increases the difficulty of correct diagnosis and treatment.

It is stressed that the disease is not always preceded by evident exposure to tick bite [2]. It is possible, however, that the bite, coinciding with a decrease in the patient's immunity, could have triggered a response of spirochetes present in her organism. It is also worth remembering that the inflammation vectors in borreliosis could be tick nymphal forms that, being very small, could have passed undetected by the patient, especially that the tick saliva has an anaesthetic properties.

In borreliosis, the pathological changes localised in the nervous system result in the polymorphism of symptoms. Because Lyme disease involves the cranial nerves, it may be manifested by non-specific symptoms [16]. They include, among others, pain complaints, such as facial and dental pain, facial nerve palsy, headache, pain in the temporomandibular joint and masticatory muscles. The symptoms in the masticatory organ at the chronic stage of borreliosis include: numbness in the tongue, especially at its tip, or in the lips, taste disorders, sore throat, toothache, tics of facial and other muscles, and headache most commonly located in the occipital area. A 4-week cycle of the symptoms is typical: 2-weeks of improvement, 2-weeks of exacerbation [4, 5]. The above-mentioned symptoms were present in the described case. Therefore, differential diagnostics is necessary and must be achieved based on clinical experience, unbiased observation and probability [2, 3]. It was partially undertaken during specialist consultations that included, among others, a dentist.

This study confirmed that multiorgan clinical manifestations of borreliosis might occur in the masticatory organ. This fact necessitates cooperation in the differential diagnosis of pain between medical and dental practitioners to ensure the best possible diagnosis and treatment.

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