

Temporomandibular joint dysfunction in a patient with Lyme disease

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ABSTRACT

Borreliosis (Lyme disease) is a chronic multisystem infectious disease transmitted by Ixodes ticks. The etiological agent of borreliosis is the spirochete *Borrelia burgdorferi*. The symptoms of borreliosis can vary depending on the attacked system and length of infection. Diagnosis is based mainly on clinical symptoms and immunoserological tests. The clinical symptoms of borreliosis also include dysfunction in the tissues and organs of the stomatognathic system.

This paper describes a case of a female patient who had experienced a long-term dysfunction of the temporomandibular joint (TMJ). After careful analysis, the symptoms were found to be caused by a chronic form of neuroborreliosis with exacerbations. The findings in this case study show that borreliosis should be considered by doctors and dentists in the differential diagnosis of pain in the area of the TMJ.

Keywords: Lyme disease; neuroborreliosis; stomatognathic system; temporomandibular joint.

INTRODUCTION

Borreliosis is a multisystem infectious disease caused by Gram-negative bacteria, strains of *Borrelia burgdorferi* sensu lato (*B. burgdorferi*): *Borrelia burgdorferi* sensu stricto, *Borrelia afzelii*, and *Borrelia garinii*. Wild rodents, wild game, and domestic animals constitute a reservoir of spirochetes, and *Ixodes ricinus* ticks serve as vectors [1, 2, 3]. The incidence of borreliosis has shown an upward trend in recent years, attributed to an invasion of ticks, a broad range of natural hosts, the role of birds in spreading the disease, and a more effective diagnosis of the disease [4].

Borreliosis is characterized by a great diversity and variability in clinical symptoms [1]. The clinical picture of borreliosis consists of dermatological, neurological, joint and cardiac symptoms [5, 6, 7, 8]. The course of the disease is long-term and the incubation period ranges 3–30 days. There are 3 stages of borreliosis, the 1st includes a characteristic erythema migrans formed at the site of the tick bite which may be associated with flu-like symptoms: fever or low-grade fever, headaches, joint and muscle pain, swollen lymph nodes and neck stiffness. Erythema migrans in untreated patients disappears after 3–4 weeks. In treated patients, erythema disappears after a few days often leaving a characteristic pigmentation or discoloration of the skin [3, 9].

In approx. 60% of untreated patients, borreliosis goes into the 2nd stage after several weeks, with the characteristic dermatological symptom of plural secondary erythema migrans, which occur regardless of the location of the tick bite. A common clinical symptom in the 2nd stage of borreliosis is acute arthritis, usually in the knee joints and, less frequently, in the

small joints of the hands and feet. The inflammatory process may also affect the temporomandibular joints (TMJs) [7, 9]. Between 15–20% of patients develop neuroborreliosis, which may be located anywhere in the nervous system [10]. This often results in radiculopathy and paralysis in cranial nerves – most often in the facial nerve. *Borrelia burgdorferi* rarely attacks the central nervous system, but in some patients, inflammation of the spinal cord or brain may be the 1st symptom of the disease [11]. Between 4–10% of patients suffer from cardiac symptoms [12].

The 3rd stage of the disease, late borreliosis, occurs a few months to several years after infection. The most common symptoms are changes in the skin (*acrodermatitis chronica atrophicans*), nervous system (peripheral and central) and the development of chronic arthritis. This stage may be associated with ischemic or hemorrhagic foci, leading to epilepsy, stroke and cognitive impairment [9].

The study was aimed at presenting the diagnostic difficulties and problems in the therapeutic management of a patient with TMJ syndrome as a result of a past cerebral stroke complicated by undiagnosed long-term neuroborreliosis.

CASE DESCRIPTION

A 71-year-old patient 1st reported to the Chair and Department of Dental Prosthetics in 2007 in order to change removable partial dentures (frameworks). An interview showed that in 1994 the patient had been hospitalized at the Department of Neurology after a stroke. The patient had been under constant scrutiny by the neurological and ophthalmic clinic because

of peripheral facial palsy on the left side and contraction of facial mimetic muscles on the left. She did not report other diseases. Since 2000, the patient had been using an upper and lower mobile dental prostheses made at an independent private dental practice. A clinical study was conducted at the Chair and Department of Dental Prosthetics by a single medical doctor (Halina Ey-Chmielewska). In the extraoral test of the facial nerve endings, the following symptoms were found: smoothing of the forehead on the left side, incompetence of the left eyelid, a drooping in the left corner of the mouth, an inconsiderable paresis of the left cheek during the inflation test and deviation of the maxilla to the left during adduction and abduction movements. An asymmetrical movement was recorded. In the palpable examination, a decreased tone of the masseter muscle on the left side was revealed. These lesions have been classified as a remnant after a past stroke. In the intraoral examination, extensive mixed deficits in the upper jaw and in the mandible, considered representative of group B4 in accordance with the Eichner classification, were noted. The mucosa of the cheeks and palate were pink and humid with no eruptions. The regions of toothless processes of the upper jaw and mandible were covered with a hard atrophic immobile mucous membrane. The attachments of the lateral frenula and the upper and lower lips were on the slopes of the alveolar processes. The prosthetic restorations used were subject to evaluation.

In the upper skeletal denture, on tooth 24, a wrought wire clasp was found to have been attached with an added tooth 25. Therefore, the upper skeletal denture had insufficient stabilization and retention on the base. The mandible revealed the atrophies of the prosthetic base within the region of the retromolar pads, which caused looseness in the lower skeletal denture and signs of damage to part of the denture's acrylic material.

A metal and ceramic crown was planned to be placed on tooth 24 in order to prepare room for clasps (the tooth has been treated endodontically under a microscope in a private dentist's office). New lower and upper removable partial dentures (frameworks) were also planned to be fitted for the patient.

In 2009, 2 years after the dental restorations, the patient reported symptoms of discomfort and tightness in the upper denture. Despite several corrections of the denture, the patient kept reporting the subjective symptom of oedema in the toothless prosthetic base within the region of the jaw tumour on the left side, the subjective symptom of compression in the upper skeletal denture on the left side and contact hypersensitivity in tooth 24. The dentists clinical study did not demonstrate any signs or symptoms of settling down in the acrylic parts of the upper denture. There were no positive signs in the horizontal and vertical percussion of tooth 24. Radiological examinations, dental panoramic radiograph and targeted imaging of tooth 24 were ordered. Despite the absence of radiological signs of periapical lesion, it was decided to extract the tooth as a potential source of discomfort, and to execute new prosthetic restorations.

Despite the removal of tooth 24 and execution of the new prosthetic restorations, the patient still complained of

subjective symptoms of periodic swelling in the prosthetic base on the left. A consultation internist was commissioned due to the occurrence of pain in the joints and muscles especially at night, accompanied by a severe headache, a slight fever of 37.8°C, and general malaise. The internist ruled out infection. Because the patient complained of pain in the cheek and the left TMJ which intensified at lower ambient temperatures, radiant toothaches on the left side which were otherwise difficult to locate, and due to the presence of earlier neurological problems, the patient was referred to a neurology clinic for further specialist diagnostics. During her stay at the neurology clinic at the Pomeranian Medical University in Szczecin (Poland), she had consultations with a neurologist, ophthalmologist, and pulmonologist. A computed tomography of the head ruled out focal lesions on the brain. The clinical and radiograph tests, as well as the magnetic resonance examination, showed no pathological changes in the TMJs. The patient was discharged in good general condition.

However, after several months, the patient's condition deteriorated. Joint and muscle pains reappeared, especially at night. This was accompanied by a severe headache, low-grade fever, and general malaise. The internist again ruled out infection. Again, clinical and radiological examinations of the teeth did not confirm any problems. The clinical examination, radiographic and magnetic resonance imaging of the TMJs showed no pathological changes.

In a targeted patient questionnaire, the patient said that in 2007 she was treated for borreliosis caused by a tick bite in the area of the left cheek while working in the garden. Twenty-four hours following the bite, erythema, and edema occurred at the site of the bite, accompanied by a peripheral paralysis of the facial nerve on the left side. She was then directed to the clinic of infectious diseases and hepatology by a family doctor, where serological tests were performed in the direction of borreliosis.

The presence of specific IgM was found in a screening test using ELISA. However, a specific Western blot test to detect spirochete specific proteins did not confirm this result. The pharmacological therapy applied doxycycline, Acyclovir, Encorton, and galvanization of the left side of the face. After partial resolution of the left facial palsy, the patient was discharged in good general condition. The borreliosis serological tests were conducted again after 4 months and came back as negative.

After this information, in 2009 a serological test in the direction of borreliosis was performed again and again, yielded negative results. Nevertheless, the patient was instructed to take the antibiotic Unidox for 4 weeks. Symptoms of discomfort subsided, i.e. the subjective sensation of edema of the prosthetic base and pressure induced by the denture on the upper left side, radiating into the area of the TMJ.

In 2010, generalized joint and muscle pain was reported by the patient. Serological tests gave a positive result, which confirmed the existence of chronic borreliosis. The patient had a consultation at the clinic of infectious diseases, where a 4-week antibiotic therapy was recommended (Duomox and Azitrox). After 4 months, further serological results were

negative. It was decided that from 2007–2010 the reported subjective feelings of discomfort and tightness of the upper denture in the area of the maxillary tuber, with radiation towards the left TMJ, could be an exacerbation of chronic neuroborreliosis, especially as after a combination of antibiotics in 2010 the feeling of discomfort subsided.

Since 2010, the patient had been under continued observation at the infectious diseases clinic and outpatient treatment of occlusal interferences.

DISCUSSION

The variety of clinical symptoms which often complicate the diagnosis of borreliosis depend on the system attacked by the spirochete. After gaining access through the skin, spirochetes may enter the lymph nodes and cause lymphadenopathy or go via the blood to different organs. According to Dworżańska and Bartosik-Psujek, the type of attack on the system depends on the morphological and antigenic variability of the spirochetes [11]. According to Wanyura et al., the genetics and immune system of the patient also play a large role here [10].

In the studied case, the difficulties in diagnosing borreliosis was caused by negative results in immunoserological tests. According to Halperin, in the first 3–6 weeks after borreliosis infection, serological test results may be negative due to the time needed for the development of antibodies in blood serum [6]. As well as this, immunoserological tests do not confirm the diagnosis on account of spirochaete antigen heterogeneity and individual variation of patient immunological response. Immunoserological tests are also ineffective in the case of seronegative borreliosis which does not induce the production of antibodies [9, 13].

As shown in the presented case, neuroborreliosis may result in a sudden occurrence of cranial neuropathies, most commonly in the facial nerve. Palsy in neuroborreliosis is usually accompanied by nonspecific symptoms in the stomatognathic system in the form of numbness in the face, radiating and hard-to-locate toothaches, headaches, as well as dysfunctions in taste and smell. Patients may also complain of sudden changes in retention and stabilization of dental restorations. Wolańska-Klimkiewicz et al. described a female patient with neuroborreliosis who experienced facial palsy and acute dental pain radiating to the eye and to the ear. The patient also complained of dryness and a metallic taste in the mouth. Pains in the stomatognathic system appeared periodically and were not caused by factors associated with the teeth [14].

In the case of unrecognized neuroborreliosis, patients are often treated by dental professionals, but this does not relieve the subjective symptoms. Heir and Fein presented the case of a patient in whom pain symptoms associated with facial nerve paralysis prompted the dentist to remove the impacted tooth as a potential cause of the ailments [15]. As recommended by Lesnicar and Zerdoner, borreliosis should be considered in the differential diagnosis of acute pain around the TMJ [7]. According to Heir and Fein, the TMJ is the 4th most likely joint to be

a site of the inflammatory process in the course of the disease. Arthroscopy of the affected TMJ revealed inflammation and swelling of the synovium [16]. "The synovial membrane becomes hypertrophic with numerous folds producing villus like structures overlaid by hyperplastic synovial cells. Varying degrees of synovial edema are present. The presence of vasculopathy within the synovium and extensive fibrin deposits in the stroma are very suggestive of Lyme arthritis, even in the absence of clinical history" [17]. In the course of borreliosis-induced inflammation of the TMJ, patients may experience a sharp pain in the ear when chewing and jaw abduction lasting about 2 weeks and repeated with characteristic cyclicity every 1–3 months [16]. There may be tenderness to palpation, and restriction of movement in the affected joint [7, 18].

The diagnosis of neuroborreliosis is a difficult clinical problem. Its rich symptomatology arises from the possibility of the disease being located anywhere in the nervous system. Symptoms may occur directly after a tick bite, during the development of the local skin lesion as in the case described above, or months or years after infection [11]. The symptoms described in our case report were typical for neuroborreliosis. The diagnosis was additionally hampered by paralytic changes in the stomatognathic system caused by a history of stroke.

CONCLUSIONS

In the case of difficult-to-diagnose subjective symptoms of dysfunctions in the area of TMJs or stomatognathic system, one should take into account the possibility of other causes, including the occurrence of borreliosis or neuroborreliosis. Full diagnosis of this disease is based on the precise analysis of the patient's medical history, clinical signs and symptoms and laboratory test results such as C-reactive protein (CRP), erythrocyte sedimentation rate (ESR) and anti-CCP rheumatoid index. The implementation of additional laboratory tests enables doctors to differentiate between chronic borreliosis and diseases of a rheumatoid background. The occurrence of unspecific symptoms, unconfirmed pains, abnormalities in the mobility of the TMJs, elevated body temperature, frequent power diminution in the morning, hemi-headaches and the symptoms of uni- or bilateral masseter muscle numbness suggests a medical history should be taken. As well as this, laboratory tests should be considered – including a specific IgM class antibody test using the ELISA screening method and specific Western-blotting to detect specific proteins of *Borrelia burgdorferi* – in order to exclude tick-borne diseases.

The dentist is often the 1st person who has the opportunity to observe the symptoms of borreliosis and neuroborreliosis within the tissues and organs of the stomatognathic system. Its early detection gives a chance for targeted therapy and the rapid implementation of antibiotics to prevent irreversible changes in organs, and against chronic arthritis, neurological complications and myocarditis. It gives the patient a chance to avoid permanent disability. A delayed or non-comprehensive therapy may lead to a transition from the acute to the subacute or

chronic stages of the disease. This, in turn, may result in the appearance of remissions or exacerbations, making it difficult to perform diagnosis and therapy. Changes located in the stomatognathic system can result in unnecessary root canal treatments, extractions and higher costs of restorative therapies.

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