

# Orofacial manifestations of chikungunya infection. Case report

## *Manifestações orofaciais após infecção por vírus da chikungunya. Relato de caso*

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### ABSTRACT

**BACKGROUND AND OBJECTIVES:** The chikungunya virus is a human pathogen responsible for a disease characterized by fever, headache, myalgia, skin rash and acute and persistent arthralgia. The purpose of this case report was to describe the orofacial manifestations of a patient infected with the chikungunya virus.

**CASE REPORT:** A female patient was referred to the Universidade Federal de Juiz de Fora, MG dental clinic due to severe facial pain. Two weeks earlier, she had been diagnosed with chikungunya virus infection by ELISA. After the febrile period and skin rash, the patient presented severe pain in the shoulders, knees, and face, which make it difficult to move and perform daily activities. She was diagnosed with temporomandibular disorders (arthralgia and myofascial pain in the masseter muscle on the right side). The patient was counseled about diet free of pain, hot packs and massages in the painful region. She was already self-medicated with corticosteroids. In addition, she was instructed to seek a specialist for her body pain. The manifestations caused by infection were healed after 10 days of the beginning of the use of corticosteroids and counseling.

**CONCLUSION:** To date, no reports have been published in the literature about the orofacial manifestation of chikungunya virus, which could serve as a basis to aid in diagnosis temporomandibular joint disorders secondary to chikungunya virus or resulting from possible psychological alteration due to constant generalized pain) and treatment. The detailed anamnesis provided information about a probable temporomandibular joint disorder secondary to Chikungunya virus infection, and it was remarkable as improvement of the systemic factors resulted in the remission of orofacial symptomatology.

**Keywords:** Chikungunya virus, Facial pain, Temporomandibular joint disorders.

### RESUMO

**JUSTIFICATIVA E OBJETIVOS:** O vírus chikungunya é um patógeno humano responsável por uma doença caracterizada por febre, dor de cabeça, mialgia, erupção cutânea e artralgia aguda e persistente. O objetivo deste relato de caso foi descrever as manifestações orofaciais de uma paciente infectada pelo vírus chikungunya.

**RELATO DO CASO:** Paciente do sexo feminino foi encaminhada para a clínica odontológica da Universidade Federal de Juiz de Fora, MG, devido à dor orofacial grave. Duas semanas antes, ela havia sido diagnosticada com infecção por vírus chikungunya. Após período febril e erupção cutânea, a paciente apresentou dor intensa nos ombros, joelhos e face, que dificultava a movimentação e realização das atividades diárias. Foi diagnosticada com distúrbios temporomandibulares (artralgia e dor miofascial com referência do músculo masseter no lado direito). A paciente foi orientada sobre dieta livre de dor, compressas quentes e massagens na região dolorosa. Ela já se automedicava com corticosteróides. Foi instruída a procurar especialista para suas dores no corpo. As manifestações provocadas pela infecção foram curadas após 10 dias do início do uso de corticosteroides e aconselhamento.

**CONCLUSÃO:** Até o momento, nenhum relato foi publicado na literatura sobre a manifestação orofacial do vírus chikungunya, que poderia servir de base para auxiliar no diagnóstico de disfunção temporomandibular secundária ao vírus chikungunya ou resultante de possível alteração psicológica por dor generalizada constante) e tratamento. A anamnese detalhada forneceu informações sobre uma provável disfunção temporomandibular secundária à infecção pelo vírus chikungunya e foi notável, pois a melhora dos fatores sistêmicos resultou na remissão do sintoma orofacial.

**Descritores:** Dor facial, Vírus chikungunya, Transtornos da articulação temporomandibular.

### INTRODUCTION

Orofacial pain can occur as a sequela of various causes, for example of infectious disease. Chikungunya virus (CHIKV) is a mosquito-transmitted virus in the Alphavirus genus in the family *Togaviridae*<sup>1</sup>, which is readily transmitted to humans by infected mosquito vectors, *Aedes aegypti* and *Aedes albopictus*<sup>2,3</sup>. It is a debilitating viral illness of global concern due to its escalating outbreaks in different parts of the world<sup>2</sup>. In the human organism, the virus causes an acute infection characterized by high fever, debilitating polyarthralgia causing intense pain and swelling, myalgia, rigor, myositis, headache, chills, fatigue, photophobia, and skin rash<sup>3,4</sup>. In addition, severe neurological and

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hemorrhagic diseases and deaths were associated with an outbreak of CHIKV<sup>3</sup>.

However, the infection is self-limiting and usually resolves within 3-4 days except for the joint symptoms that may persist for a longer period<sup>2</sup>; many patients experience recurring disabling pain for months to years<sup>5,6</sup>. The long-term chronic arthralgia and myalgia can have an enormous impact on the individual's quality of life<sup>7</sup>, and on society in terms of morbidity and economic productivity<sup>7,8</sup>.

Despite the pathological significance of CHIKV infection, the physiological and molecular mechanisms occurring during viral infection are still not well-defined<sup>2</sup>.

Thus, the objective of this study was to report a clinical case of orofacial manifestations after CHIKV infection.

## CASE REPORT

A 38 years old female came to the Dentist Department of the Juiz de Fora Federal University, Governador Valadares campus, with a complaint of severe pain on the right side of her face. About 2 months before the first dentistry visit, the patient was diagnosed with a Chikungunya virus infection, through clinical diagnosis: high fever, skin rashes, and joint pain, especially knees, shoulders, and hands. The medical history showed a diagnosis of osteoarthritis in the knees several years ago.

For the first phase of the systemic treatment, analgesic was prescribed by the physician to control pain and fever, in addition to rest. The treatment was not successful in reducing pain. She sought other physicians, and non-steroidal anti-inflammatory (NSAIDs) and muscle relaxants were prescribed. The patient reported pain improvement only during the drug effects. After remission of the acute phase, pain in the knees, especially the right, and in the shoulders remained, resulting in difficulty in locomotion and execution of activities at work.

Additionally, 10 days at the end of the acute phase, the patient reported severe pain in rest in the right ear region, upper posterior teeth on the right side, which got worse during function, with severe intensity (8/10), a burning and in stitches quality, lasting 30 minutes if she did not take medications. The orofacial pain was daily (about 3 episodes a day), and no parafunctional habits were reported.

During palpation of the right temporomandibular joint (TMJ) and masseter muscle, it was possible to reproduce the pain reported by the patient in the right region: in front of the ear and upper posterior teeth. The diagnosis was arthralgia and myofascial pain with reference (to the superior posterior teeth). As well as clinical findings of muscle pain in the left masseter, bilateral sternocleidomastoids (SCM) and bilateral trapezius.

The patient received advice and home care that included verbal and written instructions about the TMD etiology and prognosis, pain-free diet, relaxing exercises of the jaw muscles, maintain a good posture, use of reminders to avoid possible parafunctional habits, avoid stimulant substances and cervical stretching, application of a heating pad on painful masticatory muscles (for 15 minutes) followed by stretching and self-massage twice a day. The patient was instructed to look for a rheumatologist or or-

thopedist physician because the pains of the knees and shoulders were more intense (10/10) than those of the face.

After 30 days, the patient attended for reevaluation. She reported that she had already started using corticosteroids (prednisone) by herself (35mg/day). She had a global improvement and didn't complain of pain in knees, shoulders or face. She had already returned to daily activities. She also reported sleep bruxism. During palpation, there was bilateral sensitivity in the masseter, temporal and SCM, of moderate to severe intensity – no pain in TMJ. A maxillary full-coverage occlusal appliance was made of hard acrylic and was fabricated in a dental laboratory. Counseling was reinforcement.

Twenty days later, the patient returned, and she had begun the process of reducing corticosteroid intake. She didn't report pain, but during palpation, it was still perceived moderate pain in the same muscles. The splint was delivered and adjusted immediately, with instruction to wear only at night while sleeping.

After 30 days of splint use, the patient returned reporting that she is in the final phase for the complete withdrawal of the corticosteroid and with the absence of symptomatology. During palpation, the patient reported little sensitivity in the masseter and temporal muscles, with slight intensity (not familiar to the patient). Counseling has been strengthened.

## DISCUSSION

The purpose of this study was to report an unusual case of Chikungunya infection where, after the acute phase of this pathology, the patient was diagnosed with TMD. To date, no reports have been published in the literature on the orofacial manifestation of Chikungunya infection.

The Chikungunya fever is one of the epidemics around the world<sup>2</sup>. Acute chikungunya symptoms in humans appear 3-12 days after a bite by an infected mosquito<sup>8</sup>. CHIKV replicates in the skin, and disseminates to the liver, muscle, joints, lymphoid tissue and brain, presumably through the blood<sup>3,4</sup>. The symptoms of CHIKV infection in humans joint are debilitating arthralgia that affects the peripheral joints, causing intense pain and swelling<sup>1</sup>. Besides, it includes high fever, headache, myalgia, conjunctivitis, periorbital pain, erythematous rash, and petechial spots<sup>3,4</sup>. Research in animals evidenced arthritis, tenosynovitis, and myositis in CHIKV infected neonatal and adult mice<sup>9</sup>.

CHIKV is also primarily arthritogenic, and muscle tissue has been proposed to be their target<sup>3,4</sup>. Most infected individuals complain of severe joint pain which is often incapacitating<sup>5,6</sup>. The arthralgia usually occurs in more than one joint, usually symmetrical and almost any joint can be affected, particularly in peripheral joints are affected more frequently<sup>10</sup>. Synovitis or periarticular swelling has been reported in 32-95% of patients, with large joint effusions occurring in 15% of individuals infected with Chikungunya<sup>10</sup>. The inflammatory response in the joint is very similar to that in rheumatoid arthritis, with leucocyte infiltration, cytokine production, and complement activation<sup>11,12</sup>. However, there is no data about TMJ pain in literature.

A study was done on muscle biopsies of CHIKV infected patients with myositis syndrome has identified muscle satellite cells

and not muscle fibers as the primary target viral infection<sup>13</sup>. The outbreak of CHIKV in the Réunion has documented 97.7% of myositis incidence upon CHIKV infection<sup>14</sup>. In this case report, during palpation, the clinical findings were the presence of active trigger points in the masticatory muscles that reported pain in the upper posterior teeth, reproducing one of the patient's main complaints. There was no clinical evidence supporting a myositis diagnosis of masticatory muscles: the presence of edema, erythema and/or increased temperature over the muscle<sup>15</sup>. The explanations for this may be the end of the acute phase of the pathology or the effect of the drugs for the systemic symptomatology since there are no reasons for the masticatory muscles not to be affected by CHIKV infection.

The acute phase of CHIKV infection typically lasts from a few days to a couple of weeks<sup>16</sup>. The convalescent phase of CHIKV infection is associated with the resolution of fever and viremia an induction of adaptive immunity<sup>6,17</sup>. However, arthralgia and/or myalgia may persist for weeks, months or even years<sup>6,17</sup> as in this case.

About 43-75% of CHIKV-infected patients experience persistent symptoms, including fatigue and joint pain, stiffness, and swelling, for about years<sup>5</sup>. Risk factors for developing chronic joint pain after acute CHIKV infection include increased age, hypertension and disease severity during the acute stage<sup>17</sup>, besides immunosuppressed organisms<sup>18</sup>. Diabetes, cardiovascular disease, neurological disorders, and chronic pulmonary diseases are risk factors for developing severe CHIKV disease<sup>19</sup>.

The ability of CHIKV to replicate and persist in the joints and muscle tissues is not clear<sup>13</sup>. The pain led our patient to have difficult locomotion. Most working adults become disabled with loss of mobility, hand impairment, and depressive reaction, which can last for weeks to months<sup>2</sup>. This loss of mobility was one of the main complaints associated with the difficult to raise her arms reported by the patient. Besides, the patient also reported difficulty in performing the mandibular movements, which may be justified by arthralgia and myofascial pain. Pre-existing arthritis (including rheumatoid arthritis and osteoarthritis) has been associated with prolonged rheumatic symptoms after infection with Ross River virus, chikungunya virus, or the related alphavirus Pogosta virus<sup>17</sup>. Several of the shared cytokines such as tumor necrosis factor, interleukin-1, interleukin-6, and interleukin-17 are established therapeutic targets during rheumatoid arthritis, showing their importance in pathogenesis<sup>2,20</sup>. The expression of this pro-inflammatory cytokines correlates with the severity of CHIKV-induced disease in patients<sup>21</sup>. Based on these similarities, Burt, Chen, Mahalingam<sup>20</sup> suggested that arthritogenic virus infection could exacerbate pre-existing joint pathology. This pain exacerbation in the patient's knee could be explained by the presence of a previous osteoarthritis diagnose some years ago.

There is no information in the literature about CHIKV virus infection in TMJ and/or masticatory muscles. However, as CHIKV has a high ability to spread and replicate in various tissues of the human body, an infected patient may have orofacial manifestations as there is no plausible explanation that the virus does not affect the masticatory muscles and TMJ, characterizing temporomandibular disorder (TMD). In this case, we could

point that TMD was secondary to CHIKV infection, as there is a temporal pattern with infection some days before the orofacial complaints. Systematic pathophysiologic conditions, such as this infectious condition caused by CHIKV, may influence local TMDs and should generally be managed in cooperation with the patient's primary care physician or other medical specialists (15). Pre-existing conditions of pain, as happened with this patient with severe pain widely spread after the CHIKV infection, may be considered as an independent risk factor for the early onset of TMD<sup>22</sup>.

Another aspect to be analyzed regarding the TMD is the fact that their onset may be associated to possible psychological alteration present in the patient due to the long period with strong symptoms of continuous pain in several structures of her body<sup>22,23</sup>. Thus, orofacial alterations would be consequences of psychosocial factors and not due to the direct action of CHIKV in the structures of the masticatory system. CHIKV could be considered as the initiating factor that might cause the onset of TMD because it has a multifactorial etiology; this infection may have interacted with other preexisting factors<sup>15</sup>.

A critical issue associated with CHIKV infection refers to the management of patient complaints, since, there are no double-blind, placebo-controlled studies to suggest the best therapeutic approach to these cases<sup>24</sup>. Since nowadays, there are not enough studies to determine the evolution of muscle involvement associated with CHIKV, although it appears that viral myositis, in general, tends to remission<sup>24</sup>. Good responses to high doses of steroids have been reported in many cases of arthropathy<sup>25</sup>. Steroids could regulate gene function and inactivate the related proteins involving pro-inflammatory cytokines, which could attenuate inflammatory myopathy<sup>26</sup>. The patient stated that the pain only relieved after she had started taking the corticoid.

Due to the high prevalence and the cyclical appearance of the outbreak of CHIKV infection, it is necessary to control the proliferation of the infected mosquito and more studies related to the treatment. The association of corticosteroids, counseling and home therapy has proven to be effective for symptomatic remission. A detailed anamnesis and physical examination could provide information about the secondary origin of the TMD. The case emphasizes the importance of a comprehensive evaluation of a patient with preexisting pain when new symptoms arise, worsening the systemic condition. In this case, the improvement of the systemic disease (CHIKV infection) associated with counseling and splint resulted in the remission of the orofacial symptomatology.

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