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Case Report

Chronic Cheek and Tongue Biting in Tourette Syndrome: A Case Report and Short Review

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KEYWORDS

Chronic cheek biting; chronic tongue biting; morsicatio buccarum, morsicatio linguarum; tourette syndrome

ABSTRACT

Introduction: Tourette syndrome (TS) is a common neurodevelopmental disorder characterized by motor and vocal tics, often accompanied by neurobehavioral and psychiatric comorbidities such as anxiety and depression. Morsicatio buccarum (chronic cheek biting) and morsicatio linguarum (chronic tongue biting) are oral mucosal lesions primarily caused by recurrent self-inflicted trauma, frequently associated with psychological factors like anxiety or stress. While self-harming behaviors, including tongue and cheek biting, are observed in TS patients, the specific manifestation of morsicatio buccarum and linguarum as an associated feature in TS remains under-documented. This case report aims to highlight a compelling connection between morsicatio buccarum and linguarum and Tourette syndrome, particularly in the context of stress and anxiety, which can exacerbate both tics and oral parafunctional habits.
Case: A 22-year-old male with a diagnosed history of Tourette syndrome presented with white linear elevations on the bilateral buccal mucosa and rough surfaces on the lateral borders of the bilateral tongue, which he had been aware of since elementary school. Intraoral examination confirmed diagnoses of morsicatio buccarum and morsicatio linguarum.
Case Management: The Depression, Anxiety, and Stress Scale (DASS-21) questionnaire was administered to assess psychological factors, which indicated severe depression and anxiety, and moderate stress. The patient received comprehensive communication, instruction, and education regarding his conditions.
Conclusion: This case suggests that morsicatio buccarum and linguarum can be a behavioral manifestation or co-occurring condition.

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INTRODUCTION

Tourette syndrome (TS) referred to as Tourette disorder in the recently updated Diagnostic and Statistical Manual of Mental Disorders (DSM–5), is a common neurodevelopmental disorder affecting up to 1% of the population.[1] It is characterized by multiple, recurrent, rapid, and involuntary motor and vocal tics and starts in childhood.[2] This disorder was first described by the French neurologist, Georges Gilles de la Tourette, in 1885.[3] TS is often accompanied by neurobehavioral and psychiatric comorbidities, such as obsessive-compulsive disorder (OCD), attention-deficit/hyperactivity disorder (ADHD), anxiety, and depression.[4] The onset of TS symptoms typically occurs in childhood, often beginning with mild, simple motor tics, such as eye blinking or head movements. The frequency and intensity of tics can vary and tend to worsen under conditions of stress or emotional instability, generally peaking in early adolescence before potentially improving in adulthood.[3]

Morsicatio buccarum, or chronic cheek biting, is primarily caused by recurrent self-inflicted trauma to the oral mucosa, frequently associated with psychological factors such as anxiety or stress.[5] Non-keratinized epithelial tissues, including the buccal mucosa (morsicatio buccarum), labial mucosa (morsicatio labiorum), and lateral borders of the tongue (morsicatio linguarum), are more commonly affected.[6] The reported prevalence of morsicatio buccarum ranges from 0.5% to 1.12%, with a higher incidence observed in individuals experiencing elevated stress or mental health conditions. While most cases of habitual cheek biting may not necessitate specific management due to their transient nature as a response to stress, chronic or extensive lesions warrant intervention for prevention and treatment.[7]

Individuals with TS may exhibit a range of associated behavioral phenomena. These can include self-harming behaviors such as tongue and cheek biting. While tics can often be temporarily suppressed, this typically leads to an escalating sense of inner tension, ultimately resulting in the eventual expression of the tic. Tic severity and presentation commonly fluctuate in a waxing and waning pattern, generally worsening in stressful situations and improving with relaxation or focused engagement in an activity.

This case highlights an urgent clinical concern, as morsicatio buccarum and linguarum in this context represent a form of repetitive behavior, potentially falling within the spectrum of self-harming tendencies observed in TS. While a wide spectrum of repetitive behaviors and stereotypical movements are well-documented in Tourette syndrome, the specific manifestation of morsicatio buccarum and linguarum as an associated feature in TS patients remains largely under-documented and less thoroughly investigated. Existing literature provides limited insight into the precise mechanisms underlying this potential association. However, a clear correlational links between the neurobiological underpinnings of TS and the development of self-inflicted oral lesions like morsicatio buccarum and linguarum are lacking.

CASE

A 22-year-old male presented to the Dental Hospital of Universitas Trisakti with chief complaints of white linear elevations on the bilateral buccal mucosa and rough surfaces on the lateral borders of the bilateral tongue. The patient reported being aware of these conditions since elementary school. He was unaware of their etiology, denied any associated pain, and had never sought treatment for them. He stated that the lesions had neither resolved nor significantly changed in size.

The patient's general medical history was unremarkable; he reported being healthy with no history of hospitalization, known allergies to medications or food, or current treatment by a general practitioner or specialist. He denied any systemic diseases. Regarding dental history, the patient last visited a dentist several years ago for a scaling procedure. He reported brushing his teeth twice daily with fluoride toothpaste but did not use mouthwash or dental floss. The patient admitted to parafunctional habits, consciously biting his tongue and inner cheeks, and clenching/grinding his teeth when focused or experiencing stress.

Family history was non-contributory regarding similar oral lesions, though his father had a history of lung

cancer. The patient reported consuming two balanced meals daily and maintaining adequate water intake (1.5 L/day), with no regular vitamin or supplement consumption. He occasionally experienced stress related to university studies. The patient was a smoker for 12 years, consuming an estimated 5 cigarettes per day, did not consume alcohol, and rarely engaged in regular exercise. Notably, he had a diagnosed history of Tourette syndrome (TS).

Intraoral examination revealed bilateral white lesions on the buccal mucosa (Figure 1) and the lateral borders of the tongue (Figure 2). These lesions presented with diffuse borders, exhibited a consistency similar to the surrounding healthy tissue, and were non-tender. Given their characteristic appearance, which is a direct result of chronic mechanical trauma such as habitual cheek and tongue biting, these findings were diagnosed as morsicatio buccarum with linea alba buccalis and morsicatio linguarum. Both conditions are inherently hyperkeratotic processes, reflecting an epithelial hyperplastic response to repetitive irritation, leading to increased keratin production and a characteristic rough, white surface.

(A) (B)

Figure 1. Morsicatio buccarum with linea alba buccalis on the buccal mucosa (A) Right buccal mucosa (B) Left buccal mucosa

(A) (B)

Figure 2. Morsicatio linguarum on the lateral border of the tongue (A) Left (B) Right

CASE MANAGEMENT

Further diagnostic assessment included the administration of the Depression, Anxiety, and Stress Scale (DASS-21) questionnaire. The results indicated that the patient experienced severe depression and anxiety, and moderate stress. The DASS-21 is a widely utilized self-assessment scale designed to quantify the severity of negative emotional states, including depression, anxiety, and stress.[7] While not serving as a definitive diagnostic tool for mental illness, it is valuable for assessing the intensity of an individual's mental health state.[8]

Based on the anamnesis, extraoral, and intraoral examinations, the patient was diagnosed with morsicatio buccarum with linea alba buccalis and morsicatio linguarum. The patient subsequently received comprehensive communication, instruction, and education. Symptomatic treatment was not initiated as the patient did not report any associated pain. For communication, the patient was reassured that the diagnosed conditions were harmless, non-malignant, and non-contagious variants, alleviating any concerns. Subsequently, the patient was instructed to cease the habits of cheek and tongue biting. The principal treatment for traumatic oral lesions involves the elimination of the causative source. In this case, the lesions resulted from chronic biting of the cheek and tongue, thus the primary treatment strategy focused on behavioral modification to discontinue these parafunctional habits. Furthermore, the patient was directed to manage his anxiety levels. Additionally, the use of a soft splint was recommended as a physical barrier to prevent severity of oral lesion due to habitual cheek and tongue biting, particularly during periods of unconscious engagement (e.g., during sleep or intense focus). The patient was also advised to maintain good oral hygiene and schedule routine dental check-ups every six months. Lastly, for education, the patient was educated that morsicatio buccarum and morsicatio linguarum are common findings resulting from chronic self-inflicted trauma, such as habitual cheek or tongue biting. It was explained that these conditions generally do not cause pain, but a rough sensation or discomfort may arise from chronic trauma and subsequent inflammation.

The patient was also informed that these conditions were attributed to environmental factors, specifically his conscious habit of biting his cheeks, exacerbated by predisposing psychological factors like stress or his Tourette syndrome (TS).

DISCUSSION

The morsicatio buccarum observed in this patient aligns with epidemiological data, indicating a significant prevalence in the 20-24 year old age group (1.20%), with a 43% occurrence in males.[9] Habitual cheek or tongue biting leads to chronic mechanical trauma on the superficial epithelium, clinically manifesting as rough white fragments accompanied by erythematous areas.[10] The underlying pathophysiology involves an epithelial hyperplastic response to this repetitive trauma, stimulating increased keratin production.[11] This physiological reaction to minor, chronic irritation initially presents as pale, translucent lesions that subsequently opacify and whiten, along with a characteristic coarse mucosal surface.[7]

The majority of cheek and tongue biting cases are linked to psychological factors, particularly stress, anxiety, and depression.[12] In the current patient, anxiety emerged as a significant predisposing factor, as confirmed by objective assessment (DASS 21). Anxiety, a subjective experience of worry or fear, frequently precipitates such oral parafunctional habits.[12] The patient's reported difficulty in managing emotional stress, particularly related to academic pressures, directly correlates with the persistence and potential exacerbation of his cheek and tongue biting. This is consistent with studies demonstrating a significant relationship between stress and these habits.[12] Effective management strategies, therefore, critically involve identifying and addressing these psychological risk factors, with the primary treatment principle being the elimination of the causative traumatic habit through behavioral modification and stress management.[7,9]

Crucially, this case highlights a compelling connection between morsicatio buccarum and linguarum with Tourette syndrome (TS). TS is a neurodevelopmental disorder known for its high prevalence of neurobehavioral and psychiatric comorbidities, including anxiety disorders.[13] Indeed, anxiety disorders, notably generalized anxiety, panic episodes, and separation anxiety, are frequently associated with tic disorders and often parallel tic severity.[14] Our patient explicitly reported a history of habitual cheek and tongue biting, which he consciously engaged in during periods of focus or stress. Concurrently, his TS manifested with motor tics (e.g., repetitive eye blinking) that significantly worsened under emotional stress or anxiety.

A significant correlation exists between anxiety and TS, with anxiety disorders being a highly prevalent comorbidity in TS patients.[15] Systematic reviews and meta-analyses, indicate that the estimated prevalence of anxiety in TS patients can be as high as 53.5%.[11] This comorbidity is critical because emotional states, particularly anxiety and stress, are well-documented triggers that can exacerbate tic severity in individuals with TS. The heightened anxiety experienced by many TS patients, whether as a direct comorbidity or in response to their tics, creates a reinforcing loop where psychological distress can directly intensify the motor and vocal manifestations of the syndrome.[11]

Possible mechanisms linking psychosocial stress to tic onset or exacerbation in TS involve several neurobiological pathways as shown in figure 3. Stress can activate the hypothalamic-pituitary-adrenal (HPA) axis, increasing levels of corticotropin-releasing hormone, adrenocorticotropic hormone, and cortisol in TS patients.[16] Abnormalities in dopaminergic neurotransmission and the cortical-striatal-thalamic-cortical circuitry are also implicated. Psychosocial stress may indirectly boost dopamine release from the sympathetic nervous system, potentially enhancing autoimmune responses and contributing to tic production.[16] Furthermore, TS patients often exhibit sympathetic overactivity, manifesting as increased heart rate, body temperature, nervousness, and agitation, with studies showing enhanced cardiovascular activity during mental load. This physiological response highlights how a patient's psychological and emotional state directly influences tic activity. Evidence suggests both sympathetic and parasympathetic nervous activity modulate tic expression; for instance, peripheral sympathetic nervous system modulation, as seen in biofeedback reducing skin conductivity, can decrease tic frequency.[17] The impact of emotional disturbance on striatal activity, through visceral

input from the amygdala and insular cortex, further demonstrates how stress influences the tic-generating circuitry. This intricate interplay underscores the profound connection between psychological factors and tic manifestation.

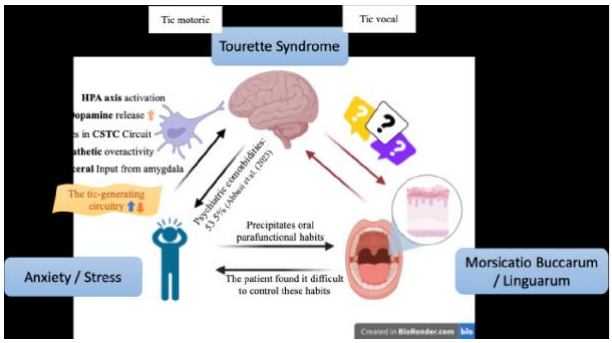

Figure 3. Interplay of Tourette syndrome, anxiety/stress, and morsicatio buccarum/linguarum

This strong correlation suggests that the patient's oral self-inflicted trauma may represent a specific manifestation within the broader spectrum of behavioral and self-harming tendencies observed in TS. While existing literature acknowledges that a subset of TS patients may exhibit self-harming behaviors like tongue and cheek biting, a clear elucidation of the precise causal or correlational links between the neurobiological underpinnings of TS and the development of morsicatio buccarum and linguarum remains largely under-documented and less thoroughly investigated. The interplay between stress, anxiety, and tic exacerbation inherent in TS appears to create a reinforcing loop, where emotional dysregulation not only drives tic frequency but also concurrently fosters or worsens the parafunctional habit of cheek and tongue biting. The patient's admitted inability to control these habits, despite discomfort, further underscores the compulsive or urge-driven nature akin to tic phenomenology.[7]

CONCLUSION

This case report provides significant clinical evidence that morsicatio buccarum and linguarum can be a behavioral manifestation or a co-occurring condition linked to Tourette syndrome. This association is particularly salient in the context of stress and anxiety, which serve as common precipitants for both tic exacerbation and oral parafunctional habits. Future research should thoroughly explore this potential neurological link, possibly through qualitative studies characterizing premonitory sensations in TS patients with oral self-injurious behaviors.

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