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Case Report

Isolated Lingual Dystonia in a Patient with Generalized Anxiety Disorder: A Case Report

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Abstract

Introduction: Lingual dystonia is a rare form of focal dystonia, and the reasons for why it occurs are not yet fully understood. Patients with this movement disorder experience incredible discomfort, distress, social embarrassment, and impaired psychological function.

Case Report: The following report discusses a 23-year-old subject who suffers from lingual dystonia and has multiple comorbidities, with an extensive drug history including various Selective Serotonin Reuptake Inhibitors (SSRIs).

Discussion: Through this case, we understand that there is a correlation between the patient's history and her dystonic attacks. There is a possible linkage between mirtazapine, a noradrenergic and specific serotonergic antidepressant (NaSSA) and other SSRIs to isolated lingual dystonia and other precipitating factors such as anxiety.

Conclusion: Isolated lingual dystonia is an extremely rare, unusual and distressing condition, of which the triggers and causes are not well understood. This case discusses the possible causes of isolated lingual dystonia and highlights the impact it can have on one's life, necessitating the need to further understand the condition.

Keywords: *Lingual Dystonia; Anxiety Disorder; Noradrenergic and Specific Serotonergic Antidepressant (NaSSA)*

Introduction

Dystonia is a Greek term that translates to altered muscle tone [1]. It is a syndrome in which specific muscle groups sustain an involuntary contractile state and can frequently be associated with twisting, repetitive movements, and abnormal postures [2,3]. Although the pathophysiology of dystonia is still being studied, one underlying theory remains consistent throughout the involvement of the basal ganglia and dopamine homeostasis (Figure 1) [3]. Dystonia can occur alongside neurological disorders, such as Wilsons, Parkinson's, and Huntington's disease, amongst many others, or in isolation [4]. For this case, isolated dystonia is the primary focus. It can be classified by etiology, site, or pattern; the first etiology group is idiopathic dystonia, which comprises two-thirds of isolated dystonia [3]. Next is the acquired or secondary dystonia, usually occurring as a byproduct of a precipitating event [5]. During birth, children can be exposed to infections, hypoxic states, or kernicterus, which can bring about the onset of childhood isolated dystonia [5].

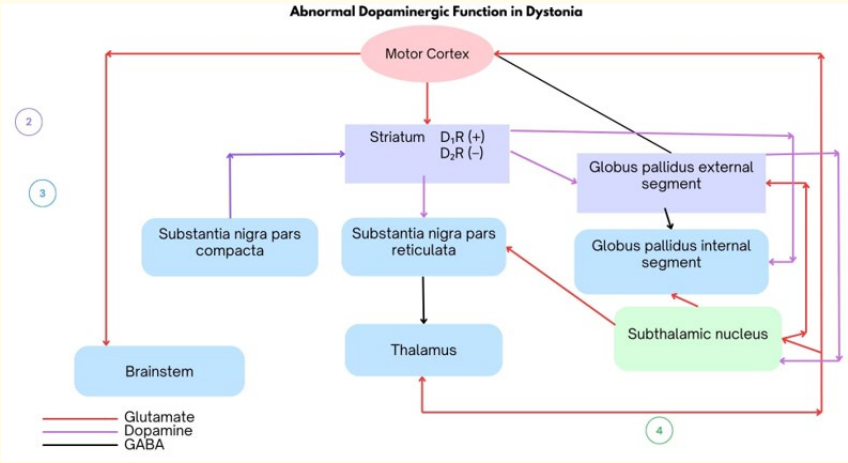

Figure 1: *Simplified schematic representation of global dopaminergic abnormalities within the basal ganglia-thalamo-cortical circuitry in dystonia. Striatal dopaminergic input from the substantia nigra, pars compacta, is weakened; dopaminergic neurotransmission is enhanced via the direct pathway and diminished via the indirect pathway.*

Furthermore, chronic exposure to drugs, most frequently dopamine antagonists causes tardive dystonia. Drugs within this class that most commonly cause tardive dystonia include antipsychotics, especially butyrophenone antipsychotics (first generation) such as haloperidol, or other drugs like metoclopramide, a dopamine antagonist used primarily to treat nausea, vomiting, and gastroparesis. These drugs put patients at risk of developing extrapyramidal side effects, such as dystonia (usually the first), akathisia, parkinsonism, and, lastly, the most chronic presentation, tardive dyskinesia [6]. Additionally, other drugs can also contribute to dystonic reactions. For instance, escitalopram, a selective serotonin reuptake inhibitor (SSRI) used to treat depression and generalized anxiety disorder (GAD), and sertraline, another SSRI used for depression and anxiety, have been associated with dystonia in rare cases. Similarly, mirtazapine has been linked to dystonia due to its effects on norepinephrine and serotonin [16]. Finally, some cases are inherited etiologically and are associated primarily with mutations in seven genes (Torsin Family 1 Member A, THAP Domain Containing 1, Guanine Nucleotide Binding Protein Alpha Activating Activity Polypeptide, Anoctamin 3, Protein Kinase, Interferon-Inducible Double-Stranded RNA Dependent Activator, Lysine Methyltransferase 2B, and Hippocalcin) [7]. The second form of classification is by the site that is affected; dystonia can affect a single part of the body, referred to as focal, or more than one part of the body, and this can be further subdivided based on the pattern; generalized (the whole/most of the body), hemi dystonia (one arm and one leg on the same side affected), segmental (adjacent parts) and multifocal (random body parts) [1]. Focal dystonia is ten times more common than generalized dystonia, of which cervical dystonia accounts for the most occurring form, spastic torticollis [1,8]. One of the rarest forms of focal dystonia, and the one seen in this case report, is lingual dystonia, often grouped with oromandibular dystonia, which refers to the stiffness of the jaw, mouth and/or tongue, or Miege's syndrome which is a combination of OMD and blepharospasm (eye dystonia) [9,10].

Lingual dystonia can be so severe that the patient has trouble swallowing, speaking, and, more concerningly, breathing [10]. Patients with this condition suffer tremendously and have impaired psychosocial function and quality of life, which unfortunately can further exacerbate the disorder [9,11]. Any unusual case that is seen in health care may be the beginning of an insightful discovery in medicine.

Therefore, it is paramount that these cases be reported and studied. This case displays a rare presentation, with no evidence of common etiology and other co-morbidities that should be questioned as the underlying cause before labelling the patient as having idiopathic dystonia. In other words, is dystonia a separate disease, or is it a consequence of the co-morbidities?

Case Report

A 23-year-old woman presented with irritability, aggressive behavior, excessive worries, sleep disturbances, anxiety, low mood, emotional distress, and feeling overwhelmed by her emotions. She has also been experiencing fatigue and struggling to manage daily activities for the past 2 years. She had been taking escitalopram for the past six months but stopped her medication two weeks ago after a short hospitalization for food poisoning. She discontinued her medication because she believed it was not controlling her episodes of tongue stiffness. However, her irritability and anxiety have worsened over the past two weeks.

The patient came to the clinic seeking further assessment and a solution for her stressful condition. Since September 2021, she has been experiencing episodes of tongue stiffness associated with panic attacks. During these panic attacks, she experiences a sudden onset of tongue protrusion and stiffness at rest, lasting for a few minutes. These episodes are accompanied by dysphagia (difficulty swallowing), speech difficulties, and breathing difficulties, occurring without any apparent triggers. The attacks are extremely distressing and intensify her anxiety.

She has been assessed by several physicians and was diagnosed with a conversion disorder (functional neurological symptom disorder). The patient's most recent medication was escitalopram. Her medication history also includes agomelatine, a melatonergic antidepressant, aripiprazole, an atypical antipsychotic, benzodiazepines, specifically clonazepam and alprazolam, which are used for managing anxiety and panic disorders, sertraline, and venlafaxine**,** Serotonin-Norepinephrine Reuptake Inhibitor (SNRI); used for major depressive disorder, generalized anxiety disorder, social anxiety disorder, and panic disorder. Over the past two years, she has been on these medications to treat her disorders. While her mood and anxiety improved with the medication, her tongue protrusion did not respond to various treatments.

There is a prior two-year history of drug abuse, which ceased before the onset of her dystonia. She used ketamine, an N-methyl-D-aspartate (NMDA) receptor antagonist, ecstasy (3,4-methylenedioxymethamphetamine) (MDMA), a serotonergic and dopaminergic stimulant, and cocaine, a dopamine reuptake inhibitor. She has had an eating disorder for the past seven years; her BMI is 17.11 kg/m^2 which classifies the patient in the underweight category (< 18.5 kg/m^2). She has excessive worries about weight gain and engages in purging behaviors followed by self-induced vomiting. She exercises excessively to lose weight; a Russell's sign was noted.

Regarding her developmental history, she comes from a middle-class family environment. She has a half-sibling from a previous parental relationship, with whom she has a competitive and jealousy-driven relationship. Family history includes an eating disorder in the half-sibling. The patient also reported a competitive dynamic for parental attention.

Given the above, the patient was referred to a neurologist for a thorough clinical assessment; she underwent multiple investigations including an Electroencephalogram (EEG), and blood tests to check for full blood count, vitamin and mineral deficiencies, infection markers, Erythrocyte Sedimentation Rate (ESR), serum drug levels and iron and copper levels. She also had a Computed Tomography (CT) scan to rule out any underlying neurological disorder. Due to no signs of abnormality from the neurological evaluation, the patient was provisionally diagnosed with idiopathic orofacial dystonia along with panic disorder, generalized anxiety disorder, and mild anorexia nervosa (purging type). An Electrocardiogram and baseline blood investigation were planned along with Mirtazapine a 30 mg, discontinuation of antipsychotics and SSRIs, psychotherapy, and long-term psychiatric and neurological follow-up.

Discussion

Lingual dystonia is a rare disease of 5-Hydroxytryptamine receptor 2 in basal ganglia or the inhibitory effect of serotonin on dopaminergic activity in the extrapyramidal pathway (Figure 2) [12,13]. The onset of her symptoms began around the same time she

began taking SSRIs, which makes these medications even more likely to be the reason for her dystonia. In addition to this, the patient's co-morbidities could also be precipitating factors. Few studies suggest mood disorders like anxiety and depression to be a cause of dystonia.

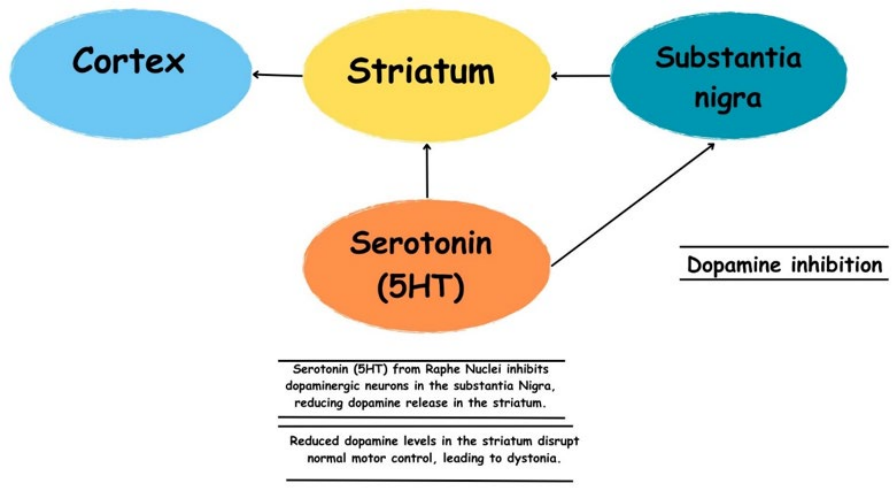

Figure 2: *Inhibitory effect of serotonin on dopaminergic activity in the extrapyramidal pathway.*

However, many corroborate a relationship between mood disorders and functional dystonia, a form of dystonia often confused with primary dystonia [14]. Interestingly, there is yet to be any strong evidence proving the association between substance misuse and dystonia. Our patient had lingual dystonia that has been labelled as idiopathic due to the unusual nature of its onset. One study proved that OMD is more frequently seen amongst the tardive dystonic group rather than idiopathic [15].

This study makes us question the extensive drug history as an account of her dystonic symptoms. Firstly, one of the primary medications this patient takes for her ongoing depression is Mirtazapine a 30 mg, which has been suggested to stimulate the substantia nigra through norepinephrine and serotonin release, thus leading to the onset of dystonia [16]. Moreover, this patient had taken two types of SSRIs, including sertraline and escitalopram, both of which have been linked to dystonia through overstimulation [13,17-19]. One study published in 2018 demonstrated that patients who have existing psychiatric illnesses and are using recreational drugs present with cervical dystonia [20]. Ketamine is known to interact with N-methyl-D-aspartate (NMDA) receptors [21]. Furthermore, ecstasy increases the net release of serotonin and possibly dopamine [22] and cocaine increases dopamine receptor activation [23], all of which have arguable participation in the development of dystonia. Despite this, our patient had stopped using substance drugs two years before the onset of her dystonia, making them unlikely to be the cause.

One major case series conducted in 1996, looked at 71 cases of SSRI-induced EPS and found that dystonia was the second most common motor dysfunction reported (28.2%). Fluoxetine was the most implicated medication associated with motor dysfunction, in 74.6% of all 71 cases [24]. Furthermore, a retrospective observational study on task-specific lingual dystonia analyzed 95 patients with a mean age of 48 years, predominantly working in high-stress occupations such as sales, customer service, and healthcare. The study found that speaking demands and work-related stress significantly contributed to the onset of dystonia. Multivariate logistic regression identified frequent speaking, high stress, and neuroleptic use as key risk factors for dystonia development. This highlights occupational and stress-related triggers as potential contributors to the condition [25]. This highlights the main associated factors studied in our case report including, SSRI's and mood/stress and other environmental factors possibly linked to dystonia. In terms of treatment options,

one notable case report included a 42-year-old woman who experienced speech-induced lingual dystonia, which was characterized by uncontrolled tongue protrusion during speech. She did not respond to anticholinergic drugs but showed significant improvement with EMG-guided botulinum toxin injections into the genioglossus muscle. This suggests that botulinum toxin can be a potential treatment when other medications fail to provide relief [26].

Conclusion

Unusual cases of isolated focal dystonia have been reported to assess and compare the possible causes of its onset, and many theories have been suggested. The possible explanations in this patient's case are endless when considering her extensive medical history. The similarities between previous reports and this report propose that health professionals should look further into the association of medications such as mirtazapine, sertraline, and escitalopram as the culprits to the onset of dystonia. As mentioned above, strong evidence proves a correlation between serotonin, norepinephrine, and basal ganglia.

Furthermore, the relationship between major mood disorders and dystonia must be reevaluated and even reformed. If anxiety and stress can exacerbate the symptoms of dystonia, what evidence do we have to suggest they have not brought on the symptoms themselves? Could this be a form of psychological dystonia, and what predisposes a patient with mood disorders to develop it? The answers to these questions are fundamental as they determine the appropriate therapeutic approach at present and for future purposes.

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