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Case Report

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Prolonged lower limb dystonia and dysphonia following general anaesthesia in a patient on Hydroxyzine

Summary

A female patient in her early 40s presented with prolonged lower limb dystonia following general anaesthesia in multiple occasions. She once developed transient aphasia and prolonged dysphonia after total intravenous anaesthesia. She had uneventful anaesthetics before the prescription of hydroxyzine for her skin condition. All post anaesthetic dystonic events were reported while she was on hydroxyzine. Spasmodic laryngeal dystonia can result in dysphonia. The possible mechanisms of dystonia can be explained using a compartmental model of the striatum. An imbalance between cholinergic and dopaminergic systems in basal ganglia results in dystonic reactions. Changes in the endocannabinoid system could partly explain the effects on movement disorders after propofol administration. Antagonistic effect of hydroxyzine on dopamine 2 receptors in the striatum can result in predominant excitatory motor effects and movement disorders.

Detailed history of the patient and analysis of findings with the clinical knowledge is important for the management of rare perioperative presentations of movement disorders.

Keywords: General anaesthesia, dystonia, dysphonia, Hydroxyzine, Propofol, movement disorder

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Introduction

Transient movement disorders are common during induction and emergence from general anaesthesia. Severe and prolonged dystonic reactions following general anaesthesia are rare, especially in patients with no other risk factors, and are frequently misinterpreted as convulsions. Laryngeal dystonia has not been reported following general anaesthesia to our knowledge. Even though hydroxyzine can rarely cause movement disorders, dystonia in patients on hydroxyzine has never previously been reported following general anaesthesia.

Case report

A female patient in her early 40s presented to us with severe sciatica pain for primary posterior fusion and decompression in the lumber region. She has a medical history of chronic pain and skin psoriasis and has undergone a total of 8 surgical procedures in the past. The patient has experienced abnormal movements in her lower limbs during most of her post-operative periods. That is explained as vigorous painful involuntary movements of lower limbs lasting for a couple of hours. The use of midazolam and magnesium has transiently reduced her signs and symptoms.

A year ago, she received total intravenous anaesthetics for a hand surgery, where she experienced aphasia for a couple of hours and dysphonia lasting for a couple of months. Dysphonia was explained as a stuttering and difficulty in controlling her voice with excessively loudness. She had no sore throat or structural anomaly on endoscopy. Her CT and MRI brain were essentially normal at that time and classified her symptoms as functional expressive dysphasia after referring to a neurologist.

She has no history of any movement disorders in the past except during the emergence of anaesthetics. She has no family history of movement disorders or problems that could relate to anaesthetics. She was on hydroxyzine for her skin condition as a long – term medication. Anaesthesia was induced with 200mg of propofol and received 60mg of rocuronium. During the 40 minutes of operative time, her vital parameters remained within normal ranges, and she was euthermic. Anaesthesia was maintained with Sevoflurane on oxygen and air. The patient received 4mg of ondansetron, 6.6mg of dexamethasone, a total of 15mg of morphine, 1g of paracetamol, 40mg of parecoxib, 1g of Magnesium,and 1mg of intrathecal morphine intraoperatively. After tracheal extubation, the patient was sent to a recovery room or a recovery department.

When she was fully awake, her legs started regular synchronous, rhythmic, and jerky movements. They were vigorous enough that her body moved or slid down the bed. The movements occurred in 4 to 5 jerks every 10 to 15 seconds. Her torso was involved in movements and abdominal and erector spinae muscles were in intermittent spasms. The patient's lower limb muscles were spastic. She received 2mg of midazolam which relieved her symptoms transiently for about 2 to 3 minutes. The patient complained of severe pain in in her lower limbs, for which she was given a further 10mgs of morphine intraveneously. No other abnormal movements or body positions were noted. She was fully conscious and cooperative throughout. Her heart rate rose to 140 beats per minute and sustained above 120 beats per minute throughout. The highest body temperature recorded was 36.8C°.

After 40 minutes of the dystonic movements, she was moved into an assisted sitting position with both legs hanging down, where complete cessation of her symptoms and signs were noted. She experienced excessive sweating and muscle pain for a couple of hours, which were treated symptomatically. The patient was discharged home on the second day, and no further abnormal movements were reported. She was feeling weak and fatigued for the next few days.

Discussion

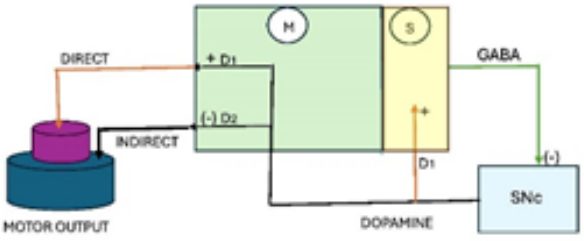
Dystonia is considered a movement disorder classically present with sustained muscle contractions and repetitive or patterned movements in varying body parts. It often produces a twisting posture and interestingly follows a task-specific movement, frequently named as 'sensory trick'. Among the other hyperkinetic movement disorders like chorea, myoclonus, akathisia, tics, and tremors, our patient's signs and symptoms tally with dystonic movement disorder. She presents with this movement disorder only after anaesthetics, therefore, it can be considered as a dystonic reaction to anaesthetics or as a complex interaction of anaesthetic with medication she is already on.

Sustained or patterned contractions of agonist and antagonistic muscles in limbs, trunk, and) neck result in involuntary movements and twisting posture. She presented with lower limb dystonia, however, she suffered from dysphonia following anaesthetics in the past, which can be a form of adductor laryngeal spasmodic dystonia resulting in voice tremor and affecting loudness of the voice.[1] Further, spasticity in the neck and facial muscles could result in life-threatening airway emergencies with upper airway obstruction. To our knowledge this is the first case report describing laryngeal dystonia following anaesthetic.

Dystonic reactions during the perioperative period are rare and mostly occur during induction and emergence, which usually be transient.[2,3] The majority of the cases of perioperative movement disorders were correlated to propofol[3] or antiemetic agents such as metoclopramide or ondansetron. In our case, ondansetron has been used without any ill effects previously. Propofol was used as the induction agent in all her previously reported dystonic events and on one occasion as total intravenous anaesthesia where severe lower limb dystonia and aphasia were reported. A clear finding of worsening severity and duration of dystonic reaction after each anaesthetic was noted in our case and such graded response following anaesthesia has been described previously.

Even though the pathophysiology of dystonia is not clearly understood, several mechanisms has been described by various authors. The classic description is the imbalance between inhibitory dopaminergic and excitatory cholinergic systems in basal ganglia. Recent works suggest that dystonia is a motor circuit disorder rather than a simple neurochemical imbalance confined to the basal ganglia. Further, studies involving tonic vibration reflex stimulus suggest that muscle spindle afferent pathways carry kinaesthetic inputs to the cerebellum.[4] Sensory inputs are directed to the basal ganglia and mainly to the stratum via the thalamus. These pathways could explain the sensory elements of dystonia.

A compartment model in the striatum could explain some of the findings related to our patient's presentation. Striosomes and matrix are considered neuro – chemically distinct compartments in the striatum. Of that, a much larger compartment, the matrix, comprises direct excitatory and indirect inhibitory neurons.[4] Excitatory outputs mediate via dopamine 1 (D1) receptors and indirect inhibitory outputs involve D2 receptors. Neurons in the striosome get inputs from the limbic system. Cholinergic neurons influence the activation of the striosomes. These striosomal neurons have inhibitory projections into substantia nigra pars compacta (SNc) which act via gamma - aminobutyric acid (GABA) receptors. Outputs from SNc have feedback into striosome and matrix mediated via dopamine. It activates predominantly the direct pathway of matrix via D1 receptors giving rise to an excitatory motor response (Figure 1).



(Three compartmental models of basal ganglia circuit. Sriosomes (S) give inhibitory projections to substantia nigra pars compacta (SNc) via gamma - aminobutyric acid (GABA) receptors. Output from SNc gives feedback into the matrix (M) via dopamine. Matrix carries direct excitatory motor outputs via dopamine 1 (D1) receptors and indirect inhibitory motor output via dopamine 2 (D2) receptors.)

Various extrapyramidal side effects have been reported concerning the use of propofol. The mechanism of such movement disorders is largely unknown but commonly relates to an imbalance of cholinergic and dopaminergic effects in basal ganglia. Propofol is known to inhibit fatty amide hydrolase, which metabolizes the endocannabinoid anandamide (AEA). This leads to a significant increase in whole brain content of AEA following propofol administration.[5] The endocannabinoid system has a fundamental role in the control of movements. Therefore, changes in the endocannabinoid system could partly explain the effects on movement disorders after propofol administration.

Hydroxyzine is a first-generation antihistamine that acts as an H1 receptor inverse agonist. It is commonly used in the treatment of itchiness, insomnia, motion sickness, and anxiety. Further, it has an antagonistic effect on D2 receptors. That could result in an inhibition of the indirect inhibitory pathway at the striatum, resulting predominance of excitatory direct motor output. There are cases of dystonia following hydroxyzine administration in the literature,[6] and involuntary motor activities reported in hydroxyzine (ATARAX™) product information. Studies have shown that long – term prescription of hydroxyzine can cause tardive dyskinesia.[7]

Our patient was on hydroxyzine for her skin condition as a long-term medication. The patient had undergone a few general anaesthetics without dystonic reactions before taking the treatment with hydroxyzine. However, since starting on hydroxyzine, she subsequently had dystonic reactions after each general anaesthetic she had undergone. It is unclear how hydroxyzine interacts with general anaesthetics, and we could not find previously published cases highlighting such interaction.

A detailed patient history and correlation of clinical findings with movement disorders are important for the diagnosis. Continued education and updated knowledge in pharmacology, medicine, and related specialties for practicing anaesthetists will improve the overall outcome in rare perioperative presentations. Early diagnosis of perioperative movement disorders will prompt specific treatments, such as anticholinergic medications, for dystonia.

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Conflicts of interest

None

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