

The Use of Smartphone Video Feature for Diagnosis and Counseling of a Propofol-Induced Acute Dystonic Reaction in a Pediatric Patient

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Abstract

The propofol-induced acute dystonic reaction is a rare postoperative complication. We report the case of a 17-year-old male with a history of acid reflux and ulcerative colitis who underwent a colonoscopy under general anesthesia and developed an acute dystonic reaction postoperatively. With his mother's written consent, his movements and physical examination were filmed on a smartphone for diagnostic and educational purposes. Total cessation of all movements occurred following administration of diphenhydramine and benztropine. The video proved useful in documenting this rare complication and in counseling the patient. With hospitals moving towards secured platforms to communicate patient information, the use of smartphones to capture a patient's disease process is now possible without violating the Health Insurance Portability and Accountability Act.

Introduction

Dystonia is a neurologic movement disorder characterized by involuntary sustained or intermittent muscle contractions resulting in abnormal and uncomfortable body movements. The term dystonia was coined in 1911 when Oppenheim used the term "dystonia musculorum deformans," to describe a rare spasm disease observed in four young patients who exhibited episodes of "hypotonic and tonic muscle spasms" [1]. Dystonic movements are typically characterized as patterned, tremulous, repetitive, and torsional, which can collectively lead to bizarre and painful posturing in patients. It is exacerbated by voluntary muscle action and associated with an overflow of muscle activation [2].

This case report investigates the occurrence of a postoperative propofol-induced acute dystonic reaction in an adolescent following general anesthesia. We discuss the utility of a smartphone to film the patient for diagnostic, educational, and counseling purposes. Written consent from the mother was obtained prior to recording. It is important to recognize the early emergence of acute dystonia secondary to anesthetic agents to ensure rapid treatment and prevent adverse complications, which can delay recovery. These complications range from mild symptoms of pain, fatigue, and anxiety, to more severe conditions of functional blindness, dysphagia, and laryngospasm. We also discuss various anesthetic medications associated with acute dystonia along with methods to treat and minimize these reactions from occurring.

Case

A 17-year-old male (62.7kg, 169cm, BMI 21.95) with a past medical history of ulcerative colitis and acid reflux presented for a colonoscopy. At the age of 16, the patient had an esophagogastroduodenoscopy (EGD) under sedation and had received midazolam 2mg, lidocaine 60mg, propofol 330mg, and fentanyl 25mcgIV. At the age of 17, he had a colonoscopy under sedation with fentanyl 30mcg, midazolam 2mg, lidocaine 60mg, propofol 200mg, and ondansetron 4mgIV without any complications. He has no known drug allergies, and current medications

are mercaptopurine 100mg every other day, mesalamine 2.4mg daily, and omeprazole 20mg three times daily. The patient has also undergone three remicade infusions. His mother has a history of Crohn's Disease, and there is no family history of neurologic disorders or epilepsy. He was born at full-term without complications and has no developmental issues. On review of systems, the patient endorsed active acid reflux, nausea, and rectal bleeding. Due to the active acid reflux and nausea, the patient was taken to the procedure room with plans for a colonoscopy under general anesthesia.

After standard ASA monitors were placed and pre-oxygenation, the patient had a rapid sequence induction with lidocaine 60mg, propofol 200mg, and succinylcholine 100mgIV. He was maintained on sevoflurane for the procedure which lasted 46 minutes and ended without complications. Soon after awakening from anesthesia, he developed periodic movements, which lasted <1 minute, but occurred repeatedly. The patient was given midazolam 2mg intravenously and was transferred to the post anesthesia care unit (PACU). While in the PACU, the patient was awake, communicating, and exhibited uncontrolled, violent jerking of his neck, left arm, and bilateral lower extremities. The first dose of midazolam appeared to be ineffective. The patient reported a lack of control but was aware of his movements. The patient was able to control his movements for a few seconds, followed by violent jerks. There was no tongue biting or incontinence. The patient was administered a repeat dose of midazolam 2mg IV immediately after arriving at the PACU and also lorazepam 1 mg IM 20 minutes later.

The patient was immediately evaluated by neurology. He had difficulty in speech due to facial muscle twitches, prolonged involuntary upward deviation of the eyes, and exhibited facial and jaw spasms, dystonic movements of his neck (from side-to-side), and abrupt jerks in the left upper extremity and bilateral lower extremities. He had a tremor with action and dysmetria on the finger-to-nose test (left worse than right). These movements were described as an extrapyramidal and oculogyric crisis. This evaluation was consistent with a diagnosis of an acute dystonic

reaction to propofol. With his mother's consent, a video of the neurologic exam was taken on a smartphone (iPhone 5, Apple Co.) using the standard video recording application provided with the phone. The 91.3 MB video was 42 seconds long with a resolution of 1080 × 1920. The exam was performed by a neurology fellow and the video was sent through the hospital's secured messaging platform, TigerText (Los Angeles, CA, USA), to the neurology attending who confirmed the diagnosis. Subsequently, the patient was given diphenhydramine 50 mg and benzotropine 2 mg IV.

He had a gradual cessation of all movements within 10 minutes. He was then admitted to the pediatric ward for observation. Post-procedure labs were within normal limits. The video was shown for educational and counseling purposes to the patient and for resident education.

Discussion and Conclusion

Dystonia is a clinical diagnosis and can be further classified as focal (one body part), segmental (2 or more contiguous body parts), multifocal (noncontiguous body parts), hemidystonic (one side of the body), or generalized [3]. During dystonic reactions, it is hypothesized that the balance between inhibitory dopamine and excitatory cholinergic receptors of the basal ganglia, which controls neuromuscular coordination, is lost [4]. Secondary dystonia as a side effect of medications is more common than primary dystonia and is often transient. This involves involuntary muscle contractions typically affecting the face, tongue, jaw, eyes, and neck, which can lead to torticollis, oculogyric crisis, retrocollis, and opisthotonus. Onset occurs rapidly and is highly disturbing to patients. Very rarely, dystonia of the larynx can occur causing life threatening laryngospasms. Patients who are most susceptible are younger, male, have a history of cocaine abuse, or a previous history of dystonic reactions.

There have been sporadic accounts in the literature regarding Crohn's Disease and neurologic complications such as headaches, peripheral neurology, seizures, and dystonia, among others [5]. In one study by Elsehety et al, 3 of 253 patients with Crohn's developed dystonia [5]. In the second study by Benavente et al, 12 patients with Crohn's out of 84 patients with inflammatory bowel disease (IBD) had neurological manifestations, of which none had dystonia [6]. While our patient has a history of Crohn's Disease, he had no prior dystonic episodes, nor any other neurologic manifestations of IBD, suggesting that his dystonic episode had an alternate etiology.

Studies have described acute dystonic reactions that occur with propofol administration during anesthesia management [7]. Propofol causes hypnosis via activation of GABA type A receptors resulting in inhibitory effects primarily in the cerebral cortex. It specifically targets postsynaptic neurons in the hippocampal CA1 region along with dopaminergic and non-dopaminergic neurons in the substantial nigra pars reticulata, which potentiate GABAergic transmission in the thalamocortical circuits. Occasionally, simultaneous excitatory effects (e.g twitching, tremor, myoclonus, and dystonic posturing) can occur due to secondary suppression of subcortical areas in the brain that control voluntary motor movement [8]. An in vivo study performed in mice demonstrated that propofol might increase motor manifestations, including opisthotonus and athetoid movements, through potentiation of agonists at glycine receptors, which behave in the CNS in an inhibitory manner. The potentiation of glycine receptor agonists can occur in subcortical areas of the brain, including the basal ganglia, which is frequently involved in movement disorders [9].

Acute dystonic reactions are not limited to propofol and can be caused by other drugs including midazolam, metoclopramide, gabapentin, trazodone, varenicline, rivastigmine, ondansetron, and numerous anesthetic agents including etomidate, thiopental, and methohexital.

Midazolam-induced abnormal movements have been reported in non-Parkinsonian individuals. The underlying mechanism is thought to be that midazolam potentiates the inhibitory effects of GABA, and anticholinergic effects in the CNS, ultimately causing extrapyramidal symptoms [10]. Ondansetron, regardless of the dose, can cause dystonic reactions. Animal studies have shown ondansetron to inhibit or reduce dopamine within the mesolimbic pathway, although the mechanism is still unclear [11]. Gabapentin induced dystonic reactions are a rare side effect, with only five cases reported in the literature [12].

Like most other agents, the cause of dystonia as a result of varying doses in comparison to a single dose, and the potential mechanism of drug induced dystonia are not known [12]. Propofol is known to cause abnormal movements ranging from myoclonic movements to dystonic and violent choreoathetotic movements in non-parkinsonian individuals that are not related to dose or speed of administration [13].

Management of acute dystonic reactions can be done in a variety of ways. Patients should be placed in a monitored setting. Initial pharmaceutical intervention includes benzotropine 1-2mg IV or diphenhydramine 50mg IV. Benzotropine is an anticholinergic drug, which works by antagonizing excess acetylcholine in the CNS. Diphenhydramine is an antihistamine with additional anticholinergic properties similar to benzotropine and is readily available for treatment due to its frequent ubiquitous use in the hospital setting. Of note, even though diphenhydramine is considered part of the initial treatment for acute dystonia, it is also known to cause acute dystonic reactions, although the underlying mechanism is unknown [14]. Prochlorperazine, benzodiazepines, and dexmedetomidine are used as alternatives to alleviate refractory acute dystonic reactions that fail with initial therapy. Benzotropine is preferred over diphenhydramine due to its shorter time to recovery and lesser side effects [4]. Propofol induced abnormal movements have been classified into either seizure-like or dystonic [11]. If the reactions are similar to seizures in nature with tonic-clonic movements than treatment includes benzodiazepines and/or other anticonvulsant medications [8].

Our case was particularly unique for several reasons. Foremost, this was the patient's first exposure to general anesthesia. His prior two procedures were performed under deep sedation. Second, he had received both ondansetron and propofol, which can both potentially cause acute dystonia. Interestingly, during his first exposure, he had received a greater accumulated dose of 300mg propofol, compared to his subsequent exposures, which were both 200mg each. Third, the patient is adolescent and male; most cases presented in the literature are female and in their mid-twenties to thirties [8]. Lastly, a video of the dystonic reaction was made to help recognize and diagnose symptoms of acute dystonia, allowing for a quicker response from physicians. This video was used at Anesthesiology and Pediatric grand rounds presentations to educate residents, nurse anesthetists, and attending staff on identifying, diagnosing and managing acute dystonic reactions. Most importantly, this video was used to help counsel the patient as he was actively going through the acute dystonic reaction. It allowed him to see his involuntary actions, and based on his movements, he was given feedback of what and why this was happening. He expressed surprise at how much he was moving and requested a copy of the video, which we sent him. This is required as the video is considered part of his medical record and HIPAA requires clinicians to provide requested medical records within 30 days.

Transmitted clinical videos are used in telemedicine for many purposes including facilitating the diagnosis of stroke [15]. While the use of this video was very helpful, caution should be exercised as it may result in a breach of HIPAA. An encrypted smart phone is one possible way to avoid a video from being obtained illicitly by a hacker. As the patient's exam included observation of his striking eye and facial movements, it was impossible to obscure his face in order to conceal his identity. Fortunately,

both he and his mother consented to its educational use. Copies of the video were only shared with clinicians and the patient himself. With the use of this video, we were able to better illustrate our exam findings in a live situation allowing for a quicker diagnosis. As a result, direct management of the dystonic reaction was provided without a delay in care. There was no patient identification information in the video.

Telemedicine can range from a two-way video communication [16] to smart phone applications displaying personal health information. All of these approaches aim to further educate the patient and/or healthcare provider with real time information. Interestingly, Applegate et al discuss a randomized pilot trial using telemedicine to perform pre-anesthesia evaluations [16]. The conclusion of the study reports higher patient satisfaction and cost saving benefits when comparing to in person visits [16]. Integrating a vast supply of data into common mobile devices such as our cellphones will play a significant role in the future. Currently, with telemedicine already used in psychiatry consults and palliative care consults to name a few, the ability to reach out to rural communities and provide affordable health care in distant locations will no longer be an issue [17].

We conclude that although acute dystonia is attributed in general anesthesia to propofol, numerous other medications have been known to cause dystonia. Early recognition, monitoring, and rapid treatment are essential to prevent adverse complications, which can delay recovery. A video can be used for rapid diagnosis, capturing rare complications, and counseling patients. It can also be used as an education tool for training residents, nurse anesthetists, and attending staff on this rare complication.

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