

Childhood dystonic reactions in the middle Black Sea region

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Abstract

Acute dystonic reactions are a worrying reason for presentation to the pediatric emergency department and the pediatric neurology clinic in childhood. It must be diagnosed and treated quickly. The aim of this study was to examine the clinical presentations, etiological factors, and prognosis of patients presenting to our regional tertiary pediatric neurology clinic with a diagnosis of acute dystonic reactions in children.

Nine pediatric patients who were treated for acute dystonic reactions between May, 2018 and May, 2020 and had adequate follow-up were included in the study. Medical record data were reviewed age, gender, etiology, features of family, treatment, and results.

Three of the patients were female and 6 were male. Their average age was 11 years (4–17). All patients were evaluated as a drug-induced acute dystonic reaction. Of the 9 patients, 5 were due to metoclopramide, 3 were due to risperidone, and 1 was due to aripiprazole. It was learned that a similar situation against other drugs developed in the family history of 3 patients. As a treatment, all of them were intramuscularly applied biperiden suitable for their weight and 30 minutes dramatic improvement was observed. Additional dose had to be administered in only 1 case. All cases were discharged for 24 hours. No problem was observed in their follow-up.

Drug-induced acute dystonic reaction can be diagnosed and has a clinical picture that completely resolves when effective treatment is applied. However, it should not be forgotten that it can reach life-threatening dimensions clinically.

Abbreviation: ODU = Ordu University.

Keywords: childhood, drug, dystonia, etiology, treatment

1. Introduction

Dystonia may occur due to an underlying disease or drug intake in susceptible individuals. The clinical picture occurs as a result of involuntary contraction of the agonist and antagonist muscles at the same time. It is a movement disorder in the form of bending, twisting, or turning in the neck, tongue, trunk, or extremities, which disturbs the patient and family.^[1]

Antihistamines, decongestants, expectorants, and mixtures containing codeine or dextromethorphan together with antipy-

retics can cause acute dystonic reactions even at high or therapeutic doses.^[2,3]

Acute onset dystonia in childhood can progress to laryngospasm and airway obstruction, so it is considered a medical emergency. It should be diagnosed quickly and treated effectively. It has often been described as case reports in the literature. To the best of our knowledge, our series is one of the largest series in the literature. In this article, 9 patients who presented to the emergency department of our hospital with acute dystonic reaction were evaluated.

2. Patients and methods

Nine patients who applied to Ordu University (ODU) Training and Research Hospital with acute dystonic reactions between May, 2018 and May, 2020 were retrospectively analyzed. All patients were questioned about their own/family history, status, additional disease, and medications were evaluated. As a treatment, biperiden was administered intramuscularly in a dose appropriate to the weight of all patients and a dramatic improvement was observed within 30 minutes. Patients who were re-evaluated clinically after 3 months were included in the study. Ethics committee approval at ODU 2020/180 was obtained for the study, and the medical records of the cases were examined. Informed consent was waived.

IBM SPSS (Statistical Package for the Social Sciences) Statistics for Windows, version 21.0, was used for analyzing data. While evaluating the study data; categorical variables were expressed as n (%), normally distributed continuous variables as mean ± standard deviation, and non-normally distributed continuous variables, as median and minimum-maximum.

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All data generated or analyzed during this study are included in this published article [and its supplementary information files].

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Table 1
Clinical findings of patients.

Number	Age	Gender	Drug	Feature
1	4	Female	Metoclopramide	None
2	8	Female	Risperidone	None
3	9	Female	Metoclopramide	She has family history
4	11	Male	Risperidone	He has family history
5	11	Female	Metoclopramide	None
6	12	Female	Aripiprazole	He has family history
7	12	Female	Risperidone	The same drug was used before
8	13	Male	Metoclopramide	Additional doze was administered
9	17	Male	Metoclopramide	The same drug was used before

3. Results

Three of the patients were girls and 6 were boys. The average age (4–17) median age was 11 years old. All patients were evaluated as a drug-induced acute dystonic reaction. Of the 9 patients, 5 were due to metoclopramide, 3 were due to risperidone, and 1 was due to aripiprazole. In cases with dystonic reactions occurring after metoclopramide, symptoms were found within the first 24 hours of drug use. In cases emerging after risperidone, the clinic occurred 1 to 3 days after the initiation of the drug. The acute dystonia case that developed after aripiprazole had manifested 48 hours after the drug was started. Clinical findings of patients are summarized in Table 1.

It was learned that a similar situation developed against other drugs in the family history of 3 patients. None of the patients had a previous clinic formed in this way. The 2 patients stated that they had used the same drugs before and that such a situation had not occurred before. These patients were those who were started on metoclopramide therapy. In the treatment, biperiden was administered intramuscularly in a dose suitable for all patients, and a dramatic improvement was observed in the patients within 30 minutes. The additional dose had to be administered in only 1 case due to metoclopramide. All cases were discharged within 24 hours. No problem was observed in the follow-up period.

4. Discussion

Dystonia is a clinical picture that occurs when the agonist and antagonist muscles contract simultaneously. It may develop after a concomitant illness or medication. The incidence of acute dystonic reaction is not currently known. Although there are different rates in the literature, its prevalence is reported as 3-30/100.000 according to the types.^[1] Within extrapyramidal symptoms, there are dystonia with a dystonic reaction, akathisia, akinesia, parkinsonism, and tardive dyskinesia.

Acute dystonic reactions usually present with uncontrollable tongue movements, opisthotonus, neck hyperextension, or torticollis. Dopamine receptor antagonist drugs often cause extrapyramidal symptoms. Mixtures containing codeine or dextromethorphan with antiemetics (metoclopramide, domperidone, and prochlorperazine), antihistamines, anesthetic agents, anti-malarial drugs, decongestants, expectorants, and antipyretics may cause an acute dystonic reaction. Metoclopramide is known to have extrapyramidal side effects such as tardive dyskinesia, parkinsonism, malignant neuroleptic syndrome, dystonia, and akathisia.^[4,5]

Acute dystonic reaction is thought to be caused by a dopaminergic and cholinergic imbalance in the basal ganglia. It has been determined that the anatomical region that causes dystonia is predominantly the basal ganglia. In particular, the sensory part of the caudate nucleus, putamen, or thalamus has a role.^[1,6]

These drug-related effects occur as idiosyncratic, not dose-dependent. Therefore, side effects may occur even at therapeutic doses. It was reported that progressive dystonia developed after therapeutic doses of phenytoin and carbamazepine in epileptic children.^[7] However, side effects occur more frequently when the recommended dose is exceeded.^[5,8,9] This condition is more common in children than in adults. The risk factors include young age, male gender, primary psychotic disorder, and previous dystonic reactions in the literature.^[10]

Dystonia is treatable and reversible when it develops due to medication. Regardless of the reason, dystonia can be easily confused with other diseases, since it is rarely seen in pediatric patients.^[11] Diagnosis of dystonia by a doctor is important in terms of rapid intervention. Because, as an acute dystonic reaction clinic, it can apply in different posture forms (Table 2).^[12,13] Additionally, hyperhidrosis, pallor, and pronounced anxiety may be observed along with spastic symptoms. Although acute dystonic reactions presented to various clinics, we most frequently encountered

Table 2
Types of acute dystonic reactions.

Type	Clinic	Patients (n)
Buccolingual crisis	Trismus, risus sardonius, dysarthria, dysphagia, grimacing, tongue protrusion	3
Oculogyric crisis	Spasm of the extraocular muscles, most commonly deviated upward	1
Torticollis crisis	Abnormal asymmetric head or neck position	5
Tortipelvic crisis	Abnormal contractions of the abdominal wall, hip, and pelvic musculature	
Opisthotonic crisis	Characteristic flexion posturing with arching of the back	
Laryngeal dystonia	Dysphonia, stridor	
Pseudomacroglossia	Patient describes the sensation of tongue swelling and protrusion	



Figure 1. A 13years old male patient with torticollis crisis.

torticollis, buccolingual, and oculogyric crises in our patients. In addition, meningitis, encephalitis, epileptic seizures, psychiatric diseases, conversion, tetanus, and metabolic disorders (hypocalcemia, hypokalemia, etc) may also cause the same clinical picture.^[4] For this reason, the patient should be evaluated in terms of metabolic disorders and comorbidities, as well as drug interrogation. Blood tests and detailed history of the patient must be taken and a differential diagnosis must be made. Figure 1 presents a dystonic reaction occurring in all of our patients, because of the well-known drugs he made in this picture, the diagnosis was easily made. In most cases, the suspicion of the medication used by the families helped us a lot in diagnosis.

When previously healthy children present with an acute dystonic reaction, medication intake should not be questioned. Apart from certain drugs that are known to cause dystonic reactions typically and frequently, even therapeutic doses of drugs that rarely cause this condition may cause this clinic. Therefore, when prescribing all drugs, risks should be considered as well as benefits, and patients and their relatives should be informed about possible side effects.^[4] Paracetamol is also a selective cyclooxygenase inhibitor and dystonic reaction is not expected at normal doses. However, it may cause such clinics as it crosses the blood-brain barrier in high doses and increases the serotonergic pathways, and disrupts the dopaminergic balance. However, it has been reported that a dystonic reaction developed in a child who received paracetamol in 4 doses of treatment in India.^[14] All of our patients had well-known agents that cause dystonic reactions in the etiology.

Central anticholinergic and antiparkinsonian drugs such as diphenhydramine, benztropine, biperiden, trihexyphenidyl, and

diazepam can be used in the treatment of acute dystonic reactions.^[1,15] After the therapeutic drugs are administered parenterally, they disappear in a short time without leaving any sequelae.^[4] However, diphenhydramine and diazepam, which we use in the treatment, can also cause acute dystonic reactions. We applied biperiden treatment, which we can easily access in our pediatric emergency, to all our patients intramuscularly at an appropriate dose and we observed complete recovery within 30 minutes. An additional dose was needed in only 1 case due to metoclopramide.

The limitation of the current study, which presented the observational data from a single center with limited sample size.

5. Conclusion

In conclusion, the acute dystonic reaction is usually a transient and treatable condition, but when misdiagnosed, it can cause delayed treatment. In the progressive case, laryngospasm can be life-threatening as it may develop. Therefore, all physicians should consider the diagnosis of drug-induced dystonic reaction and apply its treatment without delay.

Author contributions

All steps: Sevgi Çıraklı.

Data curation: Sevgi Çıraklı.

Formal analysis: Sevgi Çıraklı.

Investigation: Sevgi Çıraklı.

Methodology: Sevgi Çıraklı.

Writing – original draft: Sevgi Çıraklı.

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