CASE REPORT

Acute dystonia mimicking angioedema of the tongue: a video-illustrated case

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SUMMARY

We report a case of acute dystonia of the face, jaw and tongue caused by metoclopramide and mimicking angioedema. The patient had attacks for several years before the correct diagnosis was made and we present the first ever published video footage of an attack. This adverse drug reaction is known, but might be underdiagnosed since it can mimic a wide range of other diseases.

BACKGROUND

Acute dystonic sideeffects were very common in the past when typical antipsychotics and dopamine antagonist antiemetics were more widely used. After the introduction of atypical antipsychotics and serotonin antagonists with potent antiemetic properties the entity has become rare. The symptoms include repetitive movements, sustained muscle contractions and/or abnormal postures. Metoclopramide is a widely used neuroleptic drug usually prescribed to relieve nausea in children and adults. The drug exerts its actions through its dopamine receptor antagonistic properties hereby increasing the gastrointestinal motility and decreasing the tension of the lower gastric sphincter.¹ It is prescribed by doctors in a multitude of specialties both in private practice and in the hospital setting so it is important to raise awareness of this adverse effect.

We present a patient with acute orofacial dystonia who presented with a protruding tongue, clenched jaw and intermittent hemifacial spasms. To the best of our knowledge this is the first to be published online supplementary video footage of an episode of acute dystonia of the head and neck region due to metoclopramide treatment. This video will help medical doctors to recognise the symptoms.

CASE PRESENTATION

A 48-year-old woman was acutely referred to our ear, nose and throat department with suspected angioedema and fear of airway obstruction. The patient was fully conscious and presented with intermittent tongue protrusion and hemifacial spasms, clenched jaw and dysarthria (video 1). Her neck was uninvolved and the muscles were soft on palpation. She reported an inner restlessness described as a feeling 'in the chest' similar to anxiety, but not quite like it. The tongue was somewhat cyanotic during the episodes and seemed larger than normal, but no stridor was present and the airway was safe. During the last 4–5 h she had experienced more than 20 episodes of 1–3 min

duration. A thorough patient history revealed that she had suffered similar episodes for a year and a half. She had been examined by different medical doctors but none of them had actually seen an episode. The patient was assumed to be psychologically stressed as an explanation of the symptoms. She also suffered from migraine with nausea and had been in an accident causing whiplash symptoms. Her father had passed away from Parkinson's disease. She did not take any daily medication, but used metoclopramide intermittently during migraine episodes.

INVESTIGATIONS

The otorhinolaryngeal and neurological examinations including flexible fiberoptic laryngoscopy were normal besides the symptoms described. She had no urticaria or skin swellings. When filling out a schedule of episodes and the use of metoclopramide there was no doubt of a connection between them. The symptoms arose about 30 min after taking metoclopramide. She had never experienced episodes of dystonia or restlessness unrelated to intake of this medication. To exclude other differential diagnosis a nerve conduction examination of the muscles of the head and neck and a MRI of the cerebrum were performed with normal results.

DIFFERENTIAL DIAGNOSIS

Many differential diagnoses are important to consider in a patient such as

- ▶ Neurological disease: tics, partial/focal epilepsy, infection, Parkinson's disease;
- ▶ Angioedema without urticaria;
- ▶ Psychological disease;
- ▶ Symptoms induced by illegal drug abuse.

Video 1 Acute dystonia in a patient due to metoclopramide. The patient tries unsuccessfully to close her jaw and talk during the video.

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TREATMENT

The symptoms ceased spontaneously before the diagnosis was made, so the patient did not receive any treatment. However, anticholinergic medication would have alleviated the symptoms effectively. It is not uncommon that the acute dystonia relapse after initial treatment with anticholinergic drugs (timeframe depending on the pharmacokinetics of the causative drug), in which case the patient should return and be retreated. It is important to inform the patient about this risk.²

OUTCOME AND FOLLOW-UP

At a 2-year follow-up visit the patient has had no further episodes of dystonia or restlessness since metoclopramide was substituted with ondansetron. The restlessness described by the patient could have been a symptom of restless legs syndrome, which she could have inherited from her father, or it could have been the first sign of monogenic Parkinson's disease. The symptom, however, resolved with the dystonia and did not reappear after discontinuation of metoclopramide use. Therefore, it was considered to be akathisia, another well-known sideeffect.

DISCUSSION

Metoclopramide is a drug initially used as an antipsychotic drug because of the similarity with other selective dopamine antagonists. Nowadays the drug is widely used for treating nausea after general anaesthesia,³ during chemotherapy and in the primary sector for different kinds of gastrointestinal symptoms. The calculated risk of acute dystonia during metoclopramide treatment is approximately 0.2%, although old and very young patients might be affected with an incidence as high as 25%.⁴ There are also reports of persistent tardive dyskinesia after drug withdrawal and parkinsonism confused with regular Parkinson's disease.⁵ Few reports of laryngeal dystonia due to metoclopramide treatment have been published.⁶ With this case report we would like to raise the awareness of this rare but severe and probably underdiagnosed adverse drug reaction.

Learning points

- ▶ Metoclopramide has potential serious adverse reactions due to its antagonistic interaction with the dopamine receptor.
- ▶ The neurological symptoms include acute and tardive dyskinesia, akathisia and parkinsonism.
- ▶ The symptoms are alleviated by anticholinergic medication.
- ▶ We encourage doctors to instruct patients complaining of intermittent episodes of disease with visible signs to use a camera, for example, in a mobile phone, to record video footage or take pictures of the clinical signs.

Contributors ERR developed the idea for the case report and was responsible for contacting the patient. ERR was involved in writing of the manuscript, finding the references, revision and approval of the manuscript. KAUP was involved in critical revision of the manuscript, finding the references and final approval of the manuscript. AB was involved in critical revision and final approval of the manuscript.

Competing interests ERR taught at a conference sponsored by MSD Norway. AB has been involved in scientific research and education with CSL Behring, Jerini/Shire, Swedish Orphan Biovitrum og Viropharma.

Patient consent Obtained.

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