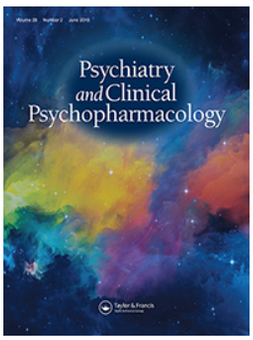

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CASE REPORT

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Psychogenic blepharospasm associated with Meige's syndrome: a case report

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ABSTRACT

Here we report a patient who presented a co-occurrence of Meige's syndrome and psychogenic blepharospasm. At the first assessments, neurologists excluded conversion disorder because of the presence of a conflict and stress, absence of any markers for Meige's syndrome, and a non-response to Botulinum toxin treatment. We determined bilateral blepharospasm, and oromandibular dystonia by neurological examination and EMG. The patient was diagnosed as primary Meige's syndrome by the neurologists. Blepharospasm, which is triggered by emotional stress, caused secondary gains against her family. We decided that the patient had both psychogenic blepharospasm and Meige's syndrome, which co-occurred nearly at the same interval three years ago. Similar to the seizure–pseudoseizure association, we supposed that Meige's syndrome and concomitant psychogenic blepharospasm may indicate a coexistence of medical and conversion symptoms as in epileptic patients.

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Blepharospasm; Meige's syndrome; conversion disorder; somatoform disorder; dystonia

Introduction

Meige's syndrome is an adult-onset idiopathic dystonic movement disorder that involves symmetrical blepharospasm, dystonia of lower facial, jaw, tongue, and neck muscles. Symptoms usually begin in the fifth or sixth decade of life with a two-fold predominance of women. Blepharospasm is the most frequent initial complaint of patients, while the other dystonic movements may gradually develop [1]. In majority of cases, Meige's syndrome develops secondary to structural lesions of the basal ganglia or the rostral brainstem [2]. Meige's syndrome has been reported to be induced by several antipsychotics, including risperidone, olanzapine, quetiapine, and aripiprazole [1,3]. It was previously reported that Botulinum toxin (BTX) injections had only a moderate improvement rates for Meige's syndrome [1]. The other treatment options in Meige's syndrome include anticholinergics, benzodiazepines, baclofen, and tetrabenazine [4].

The term psychogenic movement disorders has been applied to disorders, which are characterized by physical symptoms, specifically abnormal movements that cannot be explained by any known underlying neurological disorders [5,6]. Organic and psychogenic disorders such as epileptic and psychogenic non-epileptic seizures (PNES) can coexist in the same individual. Reports indicate that 10–58% of patients with refractory epilepsy presented PNES. 5.3–73% of patients with PNES also present epilepsy [7]. The PNES are thought to be an unintentional expression of emotional distress and can be based on learning by modelling epileptic seizures [8]. Here, we report a patient who presented a co-occurrence of Meige's syndrome and psychogenic blepharospasm.

Case presentation

Fifty-three-year-old female patient was admitted to our clinic with the symptoms of being unable to open her eyelids and contractions around her eyes, neck, and mouth which persisted for three years. Although her first EMG was reported at the upper limit of normal, she was administered a BTX injection treatment without any improvements. The medical tests for myasthenia gravis were negative. Several antidepressant treatments were also ineffective. A conversion disorder diagnosis was considered by the neurologist, since a specific organic cause could not be found. After a certain time, she has become unable to provide for her own needs and has begun to live dependent on her relatives. Blepharospasm led to embarrassment and withdrawal from social situations. The highly disabling symptoms prevented her from reading, watching television, doing everyday activities, walking, and performing other self-care tasks. Finally, the patient was admitted to our clinic to differentiate the diagnoses of Meige's syndrome and conversion disorder. In our clinical interview, her self-care was decreased, mood was depressed and her affect was anhedonic, and unhappy. Her blepharospasm and oromandibular

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dystonia were involuntary and evoked a severe anxiety. Worried and persistent thoughts about her blepharospasm were observed. There were no personal or family histories of neurological or psychiatric disorders. The patient did not receive any antipsychotics before her first symptoms emerged. Assessments including magnetic resonance imaging of the brain, EEG, serum chemistries, a complete blood count, serum ceruloplasmin, serum, and urine copper levels were within normal range. Ophthalmology examination revealed no abnormality, but bilateral blepharospasm and oromandibular dystonia were determined by neurological examination and EMG. The patient was diagnosed as primary Meige's syndrome by the neurologists. Blepharospasm Disability Index (BSDI) [9] score was 18. Initially, she was treated with 4 mg/day of biperiden. Over the two weeks, diazepam and baclofen were instituted and her symptoms were gradually improved. Her BSDI score was found to be decreased to 13 two weeks later.

In this case, stressful life events had preceded the appearance of the symptoms. Her first symptoms emerged when she experienced a financial crisis three years ago. Her all family accused her since they have seen this problem as a great fault of the patient. She reported that since she could not resist anymore to the pressure of the family, she began to close her eyes not to see their angry faces. Additionally, she told that she felt embarrassed when could not open her eyes and jaw. Blepharospasm, which is triggered by emotional stress, caused secondary gains against her family, and impaired quality of life. When her blepharospasm developed, the family did not discuss the financial crisis with the patient anymore, and they also stopped to accuse the patient. Although the patient had initially an obvious secondary gain, as the time passed she tried to get rid of her symptoms, and suffered due to failure to control her blepharospasm.

During the inpatient period, we observed that especially at her husband visit times, the patient closed her eyes, and dystonic symptoms worsened. When her husband left the unit, the contractions disappeared for more than one hour. Our patient stated that she could not control this movement, and did not know how these symptoms occurred when she met with her husband. She reported that she was bored to have this illness and was confident that her family would treat her as before the intrafamily conflicts in case she recovers from dystonia. We decided that the patient had both psychogenic blepharospasm and Meige's syndrome, which co-occurred nearly at the same interval three years ago.

Discussion

To our knowledge, this is the first case diagnosed with both Meige's syndrome and psychogenic blepharospasm. Before her symptoms emerged, the patient had experienced emotionally stressful situations which caused a threat to her ego. Blepharospasm, which is triggered by emotional stress, caused secondary gains against her family. At the first assessments, neurologists excluded conversion disorder (functional neurological disorder) due to the presence of her conflict and stress, absence of any markers for Meige's syndrome, and a non-response to BTX treatment. In our inpatient unit, the patient had persistent involuntary eye closures secondary to spasms of the orbicularis oculi and surrounding muscles. However, we noticed that blepharospasm worsened at her husband's visit times. When her husband left the clinic, she opened her eyes for a while. The patient reported that she did not know the reason of this uncontrolled dystonic movement. Neurological examination and EMG confirmed the diagnosis of Meige's syndrome. Therefore, similar to the seizure–pseudoseizure association, we supposed that Meige's syndrome and concomitant psychogenic blepharospasm may indicate a coexistence of medical and conversion symptoms as in epileptic patients.

In the case presented, a history of psychological distress and adverse life events coexisted with a true neurological disorder. Clinical differentiation of conversion disorder from Meige's syndrome has been difficult. Initially, Meige's syndrome was under-diagnosed as conversion disorder, since the first assessments excluded possible neurologic and medical conditions. Our patient exhibited a similar comorbidity pattern as seen in epilepsy–PNES association.

Conversion disorder or functional neurological symptom disorder makes up one-third of neurology outpatients and almost 9% of inpatients [10]. The DSM-IV-TR states that psychological distress is associated with the onset or exacerbation of the somatic symptom in conversion disorder [11]. ICD-10 also points out that positive signs of concurrent psychological distress are required [12]. DSM-5 reduced the number of diagnostic criteria for functional neurological symptom disorder from six to four, abandoned psychological criteria, and used psychological stressors as specifier only [13]. Traditionally, the hypothesis of conversion suggests that the symptom provides symbolic relief from an intolerable internal conflict [14]. In Freud's terms, conversion disorder is an unconscious process which the patient is unable spontaneously to make this connection. Defence mechanisms are unconscious psychological means that are beneficial in maintaining the intrapsychic balance of the individual [15]. Behavioural models suggest that conversion symptoms are learned maladaptive behaviours that are reinforced by the environment [16]. We suppose that our patient developed defence mechanisms such as closing her eyes to avoid from past painful sensations or memories. In our patient,

the blepharospasm represents the symbolic resolution of a psychological conflict and leads to avoidance of internal conflict. Emotional factors seemed to increase her vulnerability to the development of psychogenic blepharospasm and to acquire secondary gains. Our patient might also have learned that blepharospasm resulted in attention of others and fulfilled certain psychological needs.

Conclusions

This case demonstrates that the differential diagnosis of functional neurological symptom disorders per DSM-5 is a process of integrating neurological and psychosocial findings. Associated psychological stress and conflicts were related to conversion disorder, but Meige's syndrome also provided an opportunity for model learning in coexistence of both disorders [7]. We also suggest that such an association might develop in cases who are suffering from several long-term medical illnesses. Our case indicates the necessity of a close clinical collaboration of neurologists and psychiatrists in a comprehensive and ongoing psychosomatic and neurological diagnostic assessment of a neurological symptom. The absence of obvious evidences of biological dysfunction underlying neurological symptoms should not lead directly to the diagnosis of a conversion disorder. We also recommend that the clinicians should take into account the co-occurrence of Meige's syndrome and pseudo-blepharospasm in their patients.

Disclosure statement

No potential conflict of interest was reported by the authors.

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