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Apraxia of Eyelid Closure Accompanied by Denial of Eye Opening

The inability to close the eyelids voluntarily on command while retaining spontaneous blinking was first described by Roth in 1901 as a "pseudobulbar paralysis" phenomenon (1). In 1907, Lewandowsky was the first to designate this deficit as "Apraxie des Lidschlusses," an apraxia of eyelid closure (2). The loss of voluntary closure of the eyelids has been observed by other investigators, mostly in patients with diffuse, bilateral cerebral abnormalities (3-9).

In this article, we describe the clinical, evoked corneal and blink reflex, and computed tomographic (CT) findings in a patient who exhibited apraxia of eyelid closure. In addition, he also denied that his eyes remained open after attempts to close them. To our knowledge, such a deficit has not been reported before in patients with apraxia of eyelid closure.

Case Report

This 77-year-old right-handed man was admitted to our department because of an acute left-sided hemiplegia that had resulted in postural imbalance that, in turn, had led to a fall and a cerebral concussion with loss of consciousness for ~10 min and posttraumatic amnesia for a few minutes. He had hypertension, diabetes mellitus type II, and Wolf-Parkinson-White syndrome, and had experienced three previous myocardial infarctions and a number of transient ischemic attacks in the vascular area of the carotid artery.

Examination at the time of admission revealed an alert, cooperative, and normotensive man with a regular pulse rate of 75 beats/min. He was fully conscious; was normally oriented in time, place, and person; and exhibited no aphasia. His head and eyes deviated to the right. He was slightly dysarthric and had a left hemianopsia and a slight left-sided central facial paresis. The corneal reflex was normal on both sides. There was a severe left-sided hemiplegia and a left-sided hemianalgesia, of which he was aware. The deep tendon reflexes were all present. The plantar response was flexor on the right side and extensor on the left side. Two days after hospitalization, it was noticed that the patient was unable to close his eyes voluntarily on command, although they continued to blink spontaneously periodically. The corneal reflex was missing on both sides after the left cornea was touched.

When requested to close his eyes, he contracted the corrugator supercilli muscles on both sides and the procerus muscle. However, clinically there was no sign of contraction of either orbicularis oculi muscle or of relaxation of the levator palpebrae superioris muscles. On command, he was able to wink with the right eye, but not the left. Occasionally, when requested to close both eyes, he would contract either the frontalis muscles, and hence widen the eyes, or the orbicularis oculi muscle on the right side unilaterally, despite our repeated urging. There was no Bell's phenomenon. His visual threat reflex was enhanced on both sides and often resulted in forceful closure of the eyelids. However, this was not a consistent finding. Once the lids were closed passively, the patient was able to keep them closed or to open them without difficulty.

The deviation of the head and eyes was resolved. The blink rate was normal, as were the eye movements and the lid saccades in upward and downward directions. He slept with his eyes closed. His response was normal to other requests, such as opening and closing his mouth, showing his teeth, opening his eyes widely, protruding his tongue, and turning his head. Also, he demonstrated no apraxia of the limbs when following our commands, such as "comb your hair" or "brush your teeth," and his higher cerebral functions were normal.

The patient denied that his eyes remained open after his attempts to close them. Following our request that he close his eyes and the question as to whether he thought the eyes were closed, he answered, "Yes; I think so, yes; they are closed." When asked whether he could see fingers held up in front of him, he answered, "Yes, I can see them." Then, we asked him to close his eyes and to continue trying until he could not see the examiner's fingers. He contracted the same aforementioned muscles and his eyes remained open. To the question of whether his eyes were closed, he answered, "Yes; they are closed." To the question of how many fingers he saw now, he correctly answered, "Two fingers." When we told him that he should not be able to see the fingers if his eyes were actually closed, he reacted as follows: "Yes, that is curious; I cannot close my eyes; how come?" Until then, he was unaware of his inability to close his eyes. He still did not deny his left-sided hemiplegia.

The corneal and blink reflexes were evoked by techniques reported earlier (10). For eliciting the corneal reflex, a small metal sphere connected to an electronic trigger circuit was used. The blink reflex was elicited by electrical stimulation of the supraorbital nerve on both sides. A touch of the right cornea elicited a normal response in the ipsilateral orbicularis oculi muscle with a latency of 37 ms, and in the contralateral orbicularis oculi after a latent period of 43 ms, all within the normal range. To the contrary, no responses could be elicited on either side after the left cornea had been touched (Fig. 1). Stimulation of the right or left supraorbital nerve elicited a normal ipsilateral early response (R1 latency, ± 12 ms). However, no ipsilateral or contralateral late response (R2) could be recorded from the orbicularis oculi muscles after stimulation of either the right or left supraorbital nerve.

A CT brain scan performed 1 week after hospitalization showed a large, wedge-shaped infarction in the right parietal lobe, in the vascular area of the posterior branches of the right middle cerebral artery (Fig. 2). The lesion affected the lower two-thirds of the parietal lobe, extended posteriorly to the occipital lobe, and affected anteriorly the lower half of the post-central gyrus. The frontal and temporal lobes were unaffected.

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Address correspondence and reprint requests to Dr. M. Aramideh at Department of Neurology (H2-214), Academic Medical Center, Meibergdreef 9, 1105 AZ Amsterdam, The Netherlands.

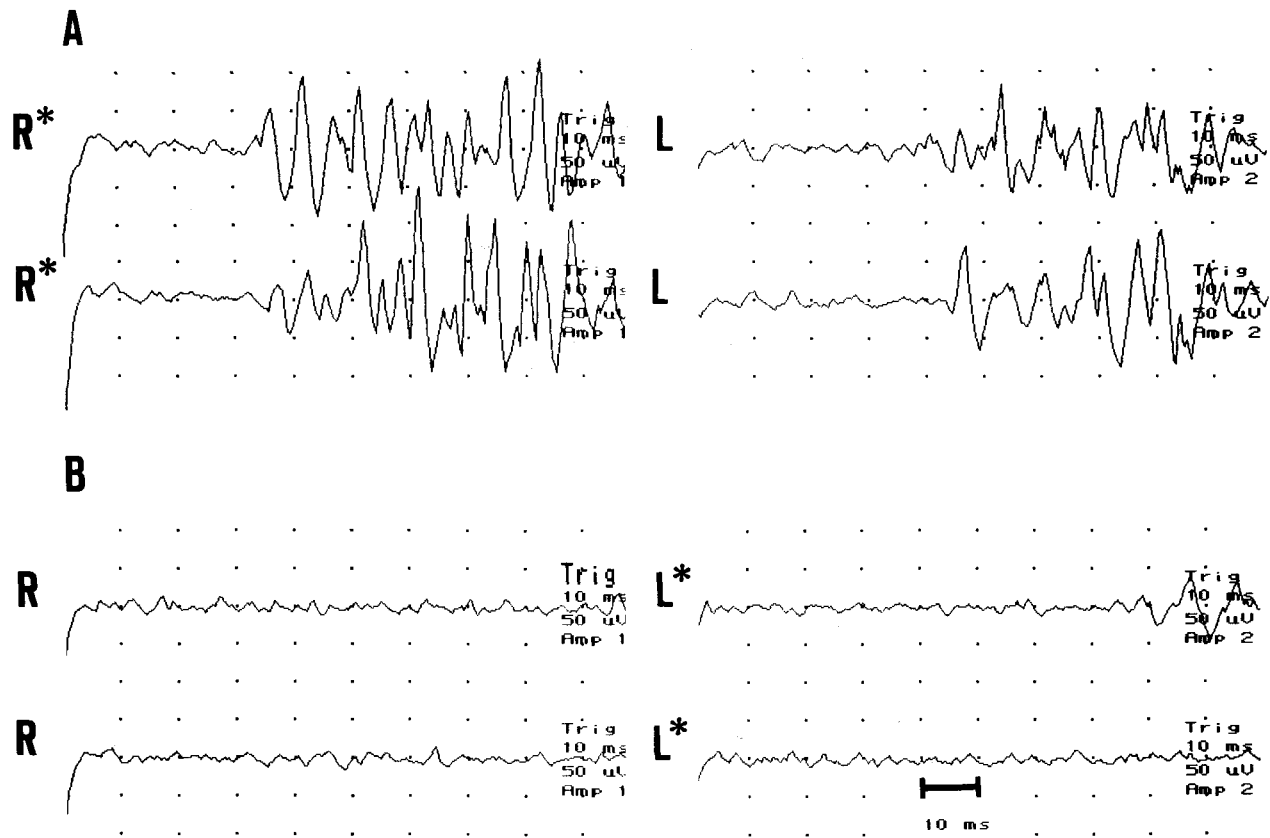


FIG. 1. A: Stimulation of the right cornea (R*, two times) elicits a normal response on both sides. B: Stimulation of the left cornea (L*, two times) elicits no response on either side.

This lesion was not seen on earlier scans that were performed several months before the ictus. Another old, small infarction, as observed on earlier scans, was seen in the middle of the left cerebellar hemisphere. Other findings were generalized cortical atrophy and slightly enlarged ventricles. No sign of intracerebral hemorrhage was noted.

Discussion

Following a vascular accident in the right hemisphere, our patient developed an apraxia in closing his eyelids and denied that his eyes remained open when he had attempted to close them. Most of the patients reported in the literature who could not voluntarily close their eyes but retained spontaneous blinking appeared to have a bilateral cortical lesion. The three patients reported by Ross Russell (6) had Creutzfeldt-Jakob disease. According to this author, the site of the lesion is probably in the cortical neurons rather than in the descending motor fibers. Lessel (5) reported two cases: one patient with vascular lesions involving both frontal lobes, and the other with amyotrophic lateral sclerosis with evidence of bilateral frontal disease. In other cases reported by Golbe et al. (8) and Jankovic (7), the patients had progressive supranuclear palsy. In one case recently reported by Abe et al. (9), the patient had neuronal loss in the precentral gyri and frontal lobes. It must be emphasized

that the presence of a small lesion undetectable by CT cannot be excluded in our patient's case.

Apraxia of eyelid closure because of a lesion in the right hemisphere in a right-handed person, as in our patient, has also been reported by Lewandowsky (2) in a patient with a sudden left-sided hemiparesis. This patient was also unaware that he was unable to close his eyes voluntarily. An associated denial that the eyes remained open on attempts to close them, as in the case in our patient, was not mentioned.

According to Liepmann (11), lesions causing apraxia would be found in the parietal lobes, in the precentral and postcentral gyri and the foot of the first two frontal gyri, with the subjacent part of the centrum semiovale, and in the corpus callosum. Our patient had an infarct in the region of the posterior branch of the right middle cerebral artery that affected, among others, the postcentral region.

In view of the anatomic site of the lesion, another finding was the absence of the corneal reflex on either side after the stimulation of the left cornea. This finding is in accord with the observation by Ongerboer de Visser (12), who found different types of corneal reflex abnormalities after lesions of the lower one-third of the postcentral region, the fibers of which probably descend through the cerebral peduncle. With respect to postcentral influences on the corneal reflex, several authors have noted that fibers from the lower part of the postcentral gyrus project to the spinal trigeminal complex (13,14). If these pro-

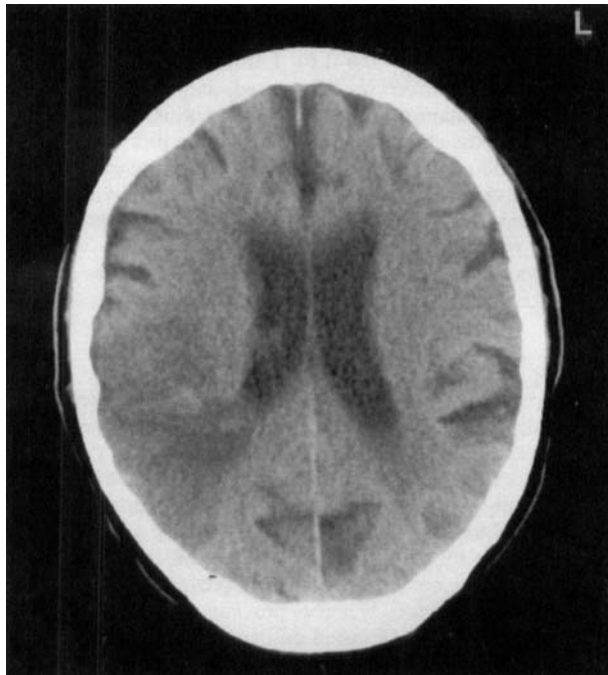


FIG. 2. Computed tomographic scan demonstrating a wedge-shaped hypodense area in the right posterior parietal region, extending to the temporoparietal region, indicative of a large infarction in the area of the posterior branches of the middle cerebral artery.

jections are involved in the corneal and blink reflexes, a lesion involving these structures might explain alterations of these reflexes in our patient and indicate that these projections exert an excitatory influence.

Despite profound abnormality of the corneal and blink reflexes, spontaneous periodic blinking was fully preserved. Apparently, these two reflexes are more sensitive to altered activity of suprasegmental structures than is spontaneous blinking.

Unfortunately, we did not perform electromyographic recordings at the orbicularis oculi and the levator palpebrae superioris muscles to acquire additional information on the origin of the failure of eyelid closure or to determine whether the reciprocal activity between these two muscles was disturbed (15,16). However, it seems that our patient's inability to close his eyes was caused by a failure in the inhibition of the tonic activity of the levator palpebrae muscles and in the contraction of the orbicularis oculi muscles.

Our patient was unable to close his lids and, furthermore, he denied that his lids remained open following his attempts to close them. Apparently, persistence of object perception does not simply result in awareness of involuntary eyelid opening and of inability to close the lids. It seems that the lesion in our patient has modified neuronal circuits in such way that his intention to close his eyes has become sufficient to create the thought that his eyes have been closed. The phenomenon of denial exhibited by our patient is similar to the denial of eye closure exhibited by patients with acute stroke, as described by Ellis and Small (17). An autopsy on one of their five patients, who denied that the eyes remained closed on attempts to open them, revealed a lesion in the frontoparietal region.

The denial of eye opening may be overlooked unless sought

by clinicians. Failure of voluntary closure of the eyelids with preservation of spontaneous blinking and denial of eye opening can be observed in patients with a unilateral parietal lobe lesion including the postcentral region.

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M. Aramideh
Department of Neurology and
Clinical Neurophysiology Unit

I. H. Kwa
J. W. M. Brans
J. D. Speelman
Department of Neurology

B. W. Ongerboer de Visser
Clinical Neurophysiology Unit

B. Verbeeten, Jr.
Department of Radiology
Academic Medical Center
Amsterdam, The Netherlands

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Tardive Dyskinesia in a Neuroleptic-Naive Patient With Bipolar-I Disorder: Persistent Exacerbation After Lithium Intoxication



Tardive or persistent dyskinesia (TD) is recognized as occurring after chronic neuroleptic treatment for schizophrenia and bipolar disorder. Although lithium is sometimes recommended as a treatment for TD (1), studies have reported that the incidence of TD does not seem to be less and may even increase when lithium is given in combination with neuroleptics (2-7). With one exception (7), however, all articles of which we are aware have reported the cases of patients who, at some time of their illness, had received neuroleptics or carbamazepine. We now report the case of a patient who had never received neuroleptics, but who, after long-term treatment with antidepressants and lithium for bipolar-I disorder, developed orofacial dyskinesia that progressed to a severe generalized dyskinesic disorder after lithium intoxication.

Case Report

A 72-year-old single Caucasian woman had been diagnosed with bipolar-I disorder when she was in her 20s. She had been treated for >20 years in our hospital with lithium maintenance therapy and doxepin or amitriptyline. Discontinuing the antidepressant and increasing lithium dosage were sufficient to control her relatively uncommon manic phases so that no neuroleptic treatment was administered. A detailed review of our hospital records and interviews with the patient and her attending physician disclosed no evidence of dopamine antagonist use, including over-the-counter medications. In 1991, the patient developed a transient orofacial dyskinesia for the first time during a depressive episode. The results of electroencephalography, Doppler sonography, electromyography, computed tomographic brain scan, and a laboratory screen including thyroid tests, antistreptolysin O, copper, and ceruloplasmin were normal. A treatment trial with tiapride was ineffective, but the dyskinesia subsided when the depressive phase waned.

In September 1994, the patient was readmitted because of ataxic gait, slurred speech, and disorientation. She was obtunded, dehydrated, and had pronounced orolingual-facial dyskinesia with a slow and slurred speech, occasional blepharospasm, and abdominal dyskinesia compromising speech and respiration. She also had chorea of the toes and fingers, a slight

intention tremor, and ataxic gait with pronounced impairment of righting reflexes. Serum lithium was measured at 1.1 mmol/L (normal range, 0.6-1.0). Lithium intoxication due to insufficient fluid intake was diagnosed, and the lithium treatment was discontinued. The ataxia and intention tremor rapidly subsided, but the chorea of the digits and the pronounced orofacial and abdominal dyskinesia persisted (despite treatment in therapeutic dosages with trihexiphenidyl, tetrabenazine, sulpiride, tiapride, and biperiden) (see the videotape). A variation in intensity was related to the patient's mental state, with increased movement disorder during depressive episodes and reduction during periods of normality or mania.

Discussion

This patient is remarkable for developing severe persistent dyskinesia in the absence of dopamine antagonists. The persistent exacerbation of TD after lithium intoxication raises the possibility of lithium as a pathogenic agent. We have been able to locate only one case in the literature of TD appearing after lithium monotherapy. In that report (7), TD also appeared with symptoms that strongly suggested lithium toxicity in a woman aged 63. Unlike our case, the TD improved markedly with lithium discontinuation. Whereas previous attempts to explain this phenomenon have focused on the lithium's augmentation of neuroleptic effect on dopaminergic and noradrenergic receptors (6), our observation suggests that lithium may have an intrinsic capacity to induce TD. Marked effects of lithium on dopaminergic and serotonergic neurotransmission, altering basal ganglia function, have been described (8). The elderly may be especially vulnerable, even at serum levels in the high-normal range (9,10). The observation that our patient's TD increased during depressive episodes and decreased during manic or euthymic episodes confirms previous reports that have shown the same pattern of fluctuation of TD in bipolar illness (11,12).

A contribution of antidepressant medication to the causation of TD in this patient cannot be excluded. Although we are not aware of confirmed reports of TD resulting from treatment with doxepin or amitriptyline (acute dystonias have been seen, however) (13), other antidepressants, especially selective serotonin reuptake inhibitors, have been implicated in the pathogenesis of TD in a number of cases (14,15).

Although the efficacy of lithium in the treatment of bipolar-I disorder is undoubted, this case recommends, in our view, vigilance in the recognition of TD, especially in the elderly and if treatment is prolonged. If patients develop abnormal movements while on lithium therapy, treatment alternatives such as carbamazepine should be considered.

Legend to the Videotape

This 72-year-old Caucasian woman has bipolar-I disorder with orofacial dyskinesia and abdominal involuntary movements. The persistent dyskinesia developed after lithium intoxication in the absence of dopamine antagonists.

A. Meyer-Lindenberg
B. Krausnick
Center for Psychiatry
School of Medicine
Justus Liebig University
Giessen, Germany

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Address correspondence and reprint requests to Dr. A. Meyer-Lindenberg at Zentrum für Psychiatrie, Justus-Liebig-Universität, Am Steg 22, 35385 Gießen, Germany.