The Detailed İnvestigation of Eyelid-Opening Apraxia after Subthalamic Nucleus Deep Brain Stimulation

Letter to Editor,

A-67-year-old right-handed male patient with akinetic rigid Parkinson's disease (PD) had undergone subthalamic nucleus deep brain stimulation (STN–DBS) surgery [Figure 1, Table 1] due to severe freezing of gait (FOG) episodes, off periods, and dyskinesias, which could not be controlled by medical therapy. The STN–DBS surgery had provided substantial improvement in the motor symptoms including gait disturbance and FOG episodes. The levodopa equivalent daily dose was reduced by 50% after the surgery. However, soon after the operation, the patient could not initiate voluntary opening of the eyelids that was compatible with apraxia of eyelid opening (ALO). The initial DBS settings were as follow; bilateral most-dorsal monopolar active contacts; 2.5 V (right), 3 V (left); 60 us (bilateral); 130 Hz (bilateral). The DBS adjustments including voltage increments and changing the localization of the bilateral active contacts did not yield any improvement in ALO, however it resulted in deterioration in motor symptoms. Besides, switching the DBS off also did not yield an improvement in ALO. Previously, in an experienced movement disorder center, bontulinum toxin (BoNT-A) injections had been performed two times (at 3-month intervals) into the pretarsal and lateral canthus region of the orbicularis oculi which had not provided a benefit. At this point, we also perform the electromyographic studies of the levator palpebrae superioris and the orbicularis oculi to understand the nature of the disturbance, however the patient did not accept the investigation. Interestingly, the patient had to use his left hand to open the lid and maintain the opening posture. However, he could not overcome ALO using his right hand and the assistance of another physician in the opening of the lid was also ineffective. The apraxia was mildly more apparent in the left eye. Of note, apraxia did not respond to levodopa therapy.

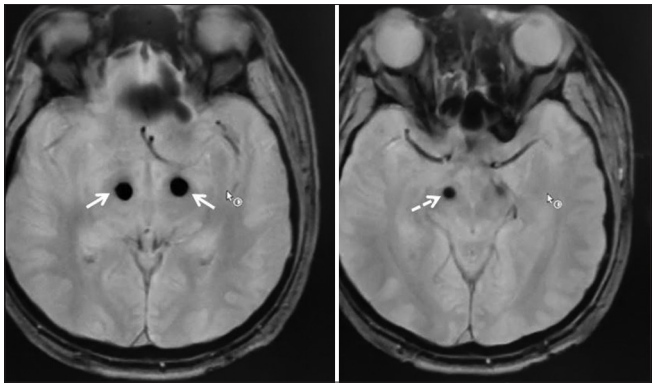

Figure 1: The cranial magnetic resonance imaging (MRI) sequences showing the bilateral electrodes of the STN–DBS. The electrode on the right side is located more inferior (jagged arrow)

The sensory tricks such as wearing goggles did not provide an amelioration. On final follow-up, 6 years after DBS, the patient still suffers from ALO [Video 1].

Discussion

On the one hand, therapeutic STN-DBS provides an effective evidence-based symptomatic relief for advanced PD.[1] On the other hand, the procedure may also result in various complications and side effects, some of which are irreversible.[1] Among them; intractable dyskinesia/dystonia, dysarthria, ALO, back pain, restless leg syndrome, and neuropsychiatric complications are the most pronounced ones.[2] Although coping with these side effects may be strictly challenging in clinical practice, the investigation of these DBS-associated neurological conditions in detail may provide crucial contributions to our current understanding of the unknown aspects of the human motor control system.

Apraxia of eyelid opening (ALO) can be defined as an inability to initiate voluntary opening of eyelids where oculomotor nerve dysfunction or ocular, myopathic or myasthenic signs do not accompany to the clinic.[3] It is a critical and common side effect following STN-DBS, in which pathophysiology and evaluation methods are not yet clear. First, the definition of ALO is under debate, as some authors do consider it as a form of focal dystonia, rather than a "true apraxia."[3] It is also discussed that ALO may be an off-phase focal dystonia possibly improving by increasing the stimulation voltages.[4] Besides, some authors suggest that ALO may be associated with a corticobulbar side effect due to lateral current spreading from the STN.[4] In our patient, the ALO was constant and did not change with alterations in the STN-DBS settings and also persisted when the stimulation was switched off. Besides, there was no amelioration with levodopa intervention. These features confirming the persistence of ALO rather suggested that the ALO in our patient might be associated with the micro-lesion effect due to the surgery, rather than a dynamic process associated with stimulation.

It is rather acknowledged that there is an abnormal supranuclear control of eyelid movement. Our patient suffered difficulty in initiating the voluntary opening of the eyelid. Remarkably, he frequently aided his left hand to initiate the opening of the lids,

Table 1: Coordinates of the tip of the electrodes relative to the mid-commissural point

	X	Y	Z	AC–PC distance
Left	−11.8	−2.0	−4.1	26.2 mm
Right	+12.0	−2.4	−3.8	

and sometimes used his hand to maintain the opening phase of the lid during gait. On the one hand, this suggests that he rather suffered activating the motor pathways in performing the onset of the eyelid movement. On the other hand, this maneuver of using his left hand to open the lid at the onset of the movement may also remind the sensory trick, an important clinical feature of dystonia. The pathophysiology of the clinic in our patient might, therefore, be affected by tactile and proprioceptive sensory inputs through the trigeminal sensory nervous system which was also hypothesized previously.[5] However, the aid of the physician's fingers did not provide an amelioration suggesting that the sensory network disturbance is not the main problem or there is certainly a disturbance also out of the sensory inputs. Possibly, the differing effect of the aid of the patient's hand may be related to the adjustment of the disturbed proprioceptive inputs (compensated by the patient's hand), rather than tactile input. However, he did not overcome the ALO by using his right hand suggesting that the activation of a network lateralized to the right hemisphere (by the movement of his left hand) may rather be processing. Although the classical knowledge associates apraxia with left hemisphere damage, recent studies have shown that the right hemisphere plays a significant role in face apraxia.[6] Of note, the electrodes were placed asymmetrically, and the right electrode was located more inferiorly [Figure 1]. The discussions regarding the lesion site due to the DBS procedure and the responsible specific networks may be further complicated. The coordination of levator palpebrae superioris and orbicularis oculi is considered to be mediated by the superior colliculus (SC).[7] Remarkably, studies on rat models revealed strong evidence regarding the connectivity between SC and rostral and dorsal sectors of the STN. We can speculate that the subtype of persistent ALO developing due to the micro-lesion effect might be associated with the affectiong of a specific region of the right STN disturbing the connections with SC. However, we can only speculate this view in the absence of related demonstrations. Future studies on these rare individuals including multiple microelectrode recording results are warranted to clarify these discussions.

In conclusion, we believe that the detailed investigation of the phenomenology in our patient may present interesting perspectives for further deliberations. The investigation of the movement disorders associated with DBS surgery is also important to enlighten the pathophysiology of many movement disorders and also provide contributions to our understanding of the human motor control system.

Abbreviations: STN subthalamic nucleus; DBS Deep brain stimulation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

Authors' contributions

Concept – Selcuk Comoglu, Halil Onder; design – Halil Onder; supervision – Selcuk Comoglu; materials – Halil Onder, Selcuk Comoglu; data collection and/or processing – Halil Onder, Bilge Kocer, Hayri Kertmen; analysis and/or interpretation – Halil Onder, Selcuk Comoglu; literature search – Halil Onder, Hayri Kertmen, Bilge Kocer; writing manuscript – Halil Onder, Selcuk Comoglu; critical review – Selcuk Comoglu, Halil Onder.

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Video available on: www.annalsofian.org

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