Reviewer's report

Title: Aggravated stuttering following subthalamic deep brain stimulation in Parkinson's disease - two case reports

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Reviewer: Harrison Walker

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The cause of stuttering is unknown. This case report describes the occurrence of neurogenic (symptomatic) stuttering and the recurrence of developmental stuttering, respectively, in two patients who underwent bilateral subthalamic deep brain stimulation (STN DBS) for advanced Parkinson's disease (PD). One of these patients had a history of a prior thalamotomy in the left cerebral hemisphere prior to DBS surgery. This report is of interest because it implicates the basal ganglia (and its connections) in the pathogenesis of stuttering and adds to prior descriptions of both worsening and improvement of stuttering following STN DBS in patients with PD.

Major compulsory revisions:

1. Was speech assessed during spontaneous speech, during reading, or both? Did singing or choral speech diminish the speech abnormality, as is common in patients with developmental stuttering?

2. Quantification of stuttering events in the various stimulation conditions with the percentage of syllables stuttered or a rating instrument such as the stuttering severity index would make it easier to interpret this manuscript. Additionally, quantification of various stuttering events (repetitions, prolongations, hesitations, etc...) would give the reader a better sense of the quality of the speech and how much the speech differed with DBS on and off.

3. Were the ratings performed by speech pathologists or personnel trained in evaluating speech disorders?

4. Were the patients truly blinded to the stimulation condition? This can be difficult, particularly when the DBS has dramatic symptomatic effects on motor function. Were efforts made to blind to the rater to tremor and other motor manifestations that might in effect unblind the rater to the stimulation condition? If not, this should be acknowledged in the manuscript.

5. Also, it would be useful to know how Parkinsonian the patients' speech was. Was there hypophonia, palilalia, or tachyphemia for instance? Did bilateral DBS worsen Parkinsonian elements of the patients' speech, independent from any effect on stuttering?

7. It would be interesting to know the handedness of the subjects with respect to the lateralization of language function. PD is an asymmetric disease in terms of its motor manifestations, and it appears that in both patients in this report, the left...
hemisphere was more severely affected than the right hemisphere by PD because the motor symptoms were worse on the right than the left.

8. These patients underwent bilateral STN DBS. Did the authors test speech with one stimulator on at a time with "motor effective" settings? It seems very possible that one of the pair of stimulators (either in the right STN or the left STN) was actually driving the speech abnormality. If the speech abnormality were present only when both stimulators were on simultaneously at "motor effective" settings, this would be of interest as well.

9. The authors conclude "stimulation of the STN itself or to structures localized in the immediate proximity" relate to the development of stuttering. Although this may be the case, the STN is one node in a complex system with direct connections to and from various anatomical structures including cerebral cortex, other basal ganglia nuclei, and brainstem structures. I think it would be more appropriate to conclude that the stimulation of the STN, its afferent or efferent projections, and/or to other structures in the vicinity of the stimulating electrode relate to the alteration of speech in these patients.

10. The authors did not cite an article with the opposite results of their findings. That case report described significant improvement in neurogenic stuttering associated with PD with unilateral left STN DBS (Walker HC et al, Journal of Speech Language and Hearing Research, 2009). The authors should explicitly discuss that there is some disagreement in the literature on this subject. How would the authors interpret their findings in the context of this result? At the least, I think the accumulated case reports argue for a role of the basal ganglia in the pathogenesis stuttering and that STN DBS appears to be able to modify it, either negatively or positively.

11. The authors seem to assume that neurogenic and developmental stuttering share the same pathogenesis. While it is certainly possible that they share some common mechanisms, the authors should acknowledge that they might in fact be different entities.

12. The authors conclude that the effect of the stimulators on motor function was clearly related to the effects on speech. Why would this necessarily be the case? Stuttering might be considered a bradykinesia of speech. If this were the case, why would DBS worsen speech yet help the arms and legs? I think the most that can be concluded is that stuttering and motor improvement in the limbs were associated with one another in these two cases - it remains possible that structures unrelated to motor function in the limbs were stimulated simultaneously at "motor effective" settings.

Minor essential revisions:

1. The authors should specify which contacts were used for monopolar stimulation on both sides of the brain.

2. The authors argue on clinical grounds that the stimulators are in the vicinity of the STN. Was post-operative MRI performed on these patients? Were the microelectrode recordings typical for the STN region?
Level of interest: An article whose findings are important to those with closely related research interests

Quality of written English: Needs some language corrections before being published

Statistical review: No, the manuscript does not need to be seen by a statistician.

Declaration of competing interests:

I declare that I have no competing interests.