Author's response to reviews

Title: When battery exhaustion lets the lame walk: A case report on the importance of long-term stimulator monitoring in deep brain stimulation

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Dear Prof Fabrizio Stocchi, dear Mr Jhonell De Los Santos

Thank you for giving us the opportunity to revise the above mentioned case report. We herewith submit a revised version with changes in red type and a revision letter highlighting the changes that we made in response to the reviewer’s comments.

We are looking forward to your evaluation of the general suitability of this revised version to the readership of BMC Neurology.

Yours sincerely,

Martin Sommer, M.D
Cover letter giving a point-by-point response to the concerns:

Reviewer 1: Bettina Debû

We thank this reviewer for the insightful comments.

The authors report on the case of a Parkinson’s disease patient whose deleterious side effects of STN stimulation (worsening of axial motor functioning, i.e. speech and gait) went unnoticed until a battery defect. Deleterious effects of STN stimulation on speech and gait in some patients are well documented and can often be related to targeting issues. Such cases are usually diagnosed in DBS expert centers, where patients’ follow-up involves a thorough assessment under the four treatment conditions (OFF med / OFF stim; OFF med / ON stim, ON med / OFF stim and ON med / ON stim). Sub-optimal selection for surgery, implantation, or parameter settings can also be suspected based on the medication dose required to improve the patients’ symptoms above STN DBS.

Thus, the case reported here is neither new nor exceptional. It illustrates patients’ management issues raised by the widespread performance of STN DBS in non-expert centers, including debatable selection criteria for surgery and lack of thorough follow-up of the patients after surgery (and possibly before surgery). It is indeed quite worrisome that the present patient’s dysarthria, at least, had not been related to STN DBS any earlier.

This case study will not bring any new information to experts in Parkinson’s disease or DBS. It might serve a didactical purpose for non-experts, but should then stress the issue of patients’ mismanagement as such.

We fully agree with this interpretation of the case report. We now stress the patients’ mismanagement in follow-up (lines 29-30 and 90) to enhance the didactic effect of the report. We specified that the surgery took place in an experienced center (line 51), but that follow-up cannot be
regarded as appropriate, and encourage re-referrals to experienced centers for systematic evaluation (line 34).

Reviewer 2: Alessandro Tessitore

We thank this reviewer for the thoughtful comments.

**Major Compulsory Revisions**

1) The case description is somehow confusing. In the abstract, the authors report a selective gait improvement only when the stimulator battery was exhausted, suggesting that was the stimulator itself the cause of clinical worsening. In the manuscript, (background and case presentation) authors state that both gait and speech improvement were related to postoperative stimulator re-programming.

Please clarify this point.

Gait and clarity of speech improved after battery exhaustion, suggesting they were caused by suboptimal stimulator settings. After a stimulation-free episode and after reprogramming of the stimulator, gait and speech continued to be improved. We tried to clarify this in the revised text (lines 45-46).

Moreover, why the authors did not stimulate more ventral contacts of the STN using a low frequency (60-80 Hz) during the stimulator re-programming phase? As demonstrated by several reports (Brozova et al., 2009, Ricchi et al., 2011; Sidiropoulos et al., 2013), this strategy might be helpful in controlling axial signs (i.e. imbalance, falls and hypophonia) of PD.

The first patient contact was in December 2008, when the above cited knowledge was not yet available. When reprogramming, we tried deeper electrode contacts which induced internal capsule stimulation and worsening of akinesia. The settings specified in lines 81-83 provided the best compromise between motor response and side effects, particularly dysarthria. In the text, we now mention strategies for optimizing stimulator settings to improve axial symptoms (lines 101-104).
2) If gait and speech were improved by battery exhaustion, it would be interesting to know why no other clinical changes were observed when the device was transiently switched off (first visit) and when the stimulator was left off. It is likely that motor fluctuations in this patient might be also related to the long disease duration, dementia or visual hallucinations. The authors should comment on this point.

Motor fluctuations were not a complaint at initial presentation and worsened significantly after four months of stimulator off, as described on line 77. This indicates effectiveness of the initial setting against motor fluctuations which then gradually returned in the stimulation off period, indicating progression of the disease. We specified this on lines 78-79.

3) Authors state that “when the battery was exhausted the patient was able to speak intelligibly with some hypophonia”. Speech problems are well known complications after DBS, especially in those patients with long disease duration and cognitive impairment. Have the authors scored speech dysfunction before and after DBS using validated scales?

Unfortunately, we did not formally score the speech dysfunction.

4) Finally, the authors state that when the battery was exhausted “rising from chair was fast, but postural responses were markedly reduced, resulting in a very unsteady gait”. However, the UPDRS motor score (24/108) did not change before and after the battery exhaustion. Authors should better clarify this point. It might helpful to show videos of the patient, if available.

Thank you for your very careful reading of the text. We checked our notes again, and as a matter of fact, gait and posture had not been scored at all before battery exhaustion, i.e. the score was 24 out of 100, and when scoring these two items at 4 out of 4 points each, 32 out of 108. We corrected this in the revised version (line 55). Unfortunately, we did not videotape the patient’s contacts.

5) This clinical case could be a good example of the complex interplay between axial motor deterioration and cognition in PD. In this patient speech, gait and postural stability worsened together with long disease duration and the appearance of visual hallucinations and dementia. Moreover, several reports (Krack et al., 2003; Funkiewicz et al., 2004; Schupbach et al., 2005;
Contarino et al., 2007; Wider et al., 2007) have highlighted that although the beneficial effects of DBS procedure are maintained at 5 years, axial motor features and cognitive decline may occur over time after implantation. Authors should comment in the discussion section the possibility that also the progression of the disease and the appearance of medication- and stimulation-resistant symptoms may play a role in this clinical case.

We now mention disease progression and occurrence of non-dopamine-responsive symptoms complicate the picture with progressing Parkinson’ disease (lines 97-99). We added the citations you raised and highlight the possible role of disease progression (lines 78-79 and 97-99).

Minor Essential Revisions

1) “patient’s wife” should be “his caregiver”

We changed this accordingly (lines 53 and 63).

2) “some hypophonia” should be “mild hypophonia”

We changed this accordingly (line 67).