DBS for dystonia

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DBS for dystonia: Should we take our patients to the swimming pool?

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Dystonia is a movement disorder defined by "continuous or intermittent muscle contractions causing abnormal, often repetitive, movements, postures, or both" [1]. Deep brain stimulation of the globus pallidus interna (DBS-GPi) is used as treatment for patients with medication-refractory dystonia and has been shown to have long-term efficacy and safety [2]. Recently, several patients with Parkinson's disease (PD) have been described who lost their ability to swim since they received subthalamic, thalamic or pallidal DBS. For this reason, we investigated the occurrence of swimming problems among our patients with dystonia who are treated by DBS. Here we describe three patients who experienced serious swimming problems since DBS-GPi implantation.

For this report, we systematically approached all dystonia patients treated by DBS-GPi in our center, through e-mail and by telephone, from December 2019 until March 2020. Of 38 patients, 14 could swim without problems, 9 had swimming problems due to their dystonia, 3 patients had not swum since DBS-implantation and there were 9 non-responders. Importantly, 3 of the 38 patients reported debilitating swimming problems that had started after DBS implantation and could not be explained by the severity of their dystonia (Table 1). Unfortunately, two of them had experienced near-drowning accidents. Before DBS implantation all three patients could swim independently (Table 1). These cases are highlighted to illustrate this worrisome problem.

Case 1. A 74-year-old man with cervical dystonia and Meige syndrome was a proficient swimmer and after his cervical dystonia was optimally managed via DBS, he picked up on swimming again. However, he could not stay afloat by way of the breaststroke; the backstroke was still possible.

A few months later, after his DBS settings had been adjusted to an interleaving program (Table 1), he was no longer able to swim at all. This led to a near-drowning accident whilst in an unfamiliar pool. His wife rescued him. When the DBS-settings were adjusted to the previous program, his swimming abilities returned, but remained clearly problematic (Supplemental Video 1).

Case 2. A 47-year-old man with idiopathic generalized dystonia swam frequently, until he developed severe worsening of dystonia. He responded exceptionally well to DBS-GPi and could resume all daily activities, except for swimming. He reported profound swimming difficulties due to inability to stay afloat, including several near-drowning experiences. Because he is unable to stay afloat for a longer period of time (Supplemental Video 2), he feels uncomfortable being close to deep water, for instance when driving a car alongside a canal.

Case 3. A 62-year-old woman with Meige syndrome was able to swim before DBS-implantation. After surgery, the blepharospasm clearly improved and she took up swimming again. However, since receiving DBS-GPi she experiences burdensome coordination issues during breaststroke. As a result she cannot keep up with her sister anymore, with whom she always swam together before DBS implantation. Furthermore, since DBS-GPi both backstroke and free stroke have become impossible for her to perform.

In this paper, we address the presence of serious swimming problems in dystonia patients receiving DBS-GPi. In several recent reports impaired swimming skills in patients with PD were described after subthalamic, thalamic and pallidal DBS [3–5]. We hypothesized that a similar mechanism could be present in dystonia. Little is known about swimming abilities and possible deterioration thereof in dystonia in general and even less in dystonia patients with DBS.

The 3 patients we describe here all developed swimming difficulties after DBS implantation, while their dystonic symptoms were stable or had improved, suggesting deterioration due to DBS implantation and not due to dystonia symptom progression.

In PD, a possible explanation for the loss of swimming skills entails disruption of signals in the supplementary motor area (SMA), a main output area of the basal ganglia [4]. Interlimb coordination has been shown to be associated to neural activity in the SMA, which might explain the coordination problems patients with DBS are facing [6]. However, other complex processes such as gait control or cycling were not affected in these PD patients [4].

Another possible explanation could be that vestibular dysfunction, described in cervical dystonia [7], contributes to the swimming problems. However, no clear vestibular symptoms such as dizziness or nystagmus were reported by the three patients. Moreover, a relationship between DBS of the ventrointermediate nucleus (VIM) and the vestibular system has been described [8], but no such relation has been described in literature for the GPi or STN.

Further research is needed to explore the relationship of swimming

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Table 1
Patient characteristics.

Case - gender	Age	Type of dystonia	Swim certificates before DBS-GPi[b]	BFMDRS[a] before DBS-GPi	BFMDRS 12 months after DBS-GPi	Current DBS-GPi settings	Relevant past DBS-GPi settings
1 - M	74	Segmental, idiopathic	A, B	28.5	30.5[c]	Left GPi 0-/3+; 4.3V; 60 μs; 135 Hz; right GPi 8-/11+; 4.3 V; 60 μs; 135 Hz	Interleaving: Left GPi 1: 0-/C+; 3.5V; 60 μs; 125 Hz; Left GPi 2: 1-/C+; 3,5V; 60 μs; 125 Hz; Right GPi 1: 8-/C+; 3,0V; 60 μs; 125 Hz; Right GPi 2: 9-/C+; 3,0V, 60 μs; 125 Hz.
2 - M	47	Generalized, idiopathic	A, B, C	85	34	Left GPi 0-/C + -; 3.2V; 90 μs, 130 Hz; right electrode off	N/A
3- F	62	Meige syndrome, idiopathic	A, B	Unknown[d]	Unknown[d]	Left GPi 2-/C+; 3.5V; 180 μs; 135 Hz; right GPi 5-/C+; 3.5V; 120 μs; 135 Hz	N/A

Note: In all three patients, postoperative neuroimaging confirmed correct position of the active electrode contacts in the dorsolateral portion of the GPi. DBS programming was performed according to our standard protocol, for which we generally select the anatomically most preferable contact for stimulation, and stimulate 0.5V below the threshold of side-effects. None of the patients had signs of parkinsonism before or after DBS implantation.

[a] BFMDRS = Burke-Fahn-Marsden Dystonia Rating Scale Movement score (range 0–120).

[b] In the Netherlands swimcertificate A comprises the minimum criteria that are set for safe swimming skills, certificate B and C cover more advanced swimming skills.

[c] For patient 1 the BFMDRS score did slightly deteriorate, but the interpretation of the BFMDRS score in this patient is hampered because he had great variability in severity of blepharospasm over the day. Furthermore, the BFMDRS is not an optimal score for measuring blepharospasm. We added the score, however, to provide a general indication of the clinical state of the patient before and after DBS.

[d] No BFMDRS scores available, but she responded well to DBS: before DBS she suffered from severe blepharospasm, dysarthria and eating difficulties due to oro-mandibular dystonia. Since DBS-implantation blepharospasm markedly improved and she has no dysarthria anymore, nor eating problems.

problems and DBS, as well as the influence of DBS on interlimb-coordination processes such as staying afloat in water.

In this report, we highlight the possibility of serious swimming problems in patients receiving DBS-GPi for dystonia, aiming to increase the awareness of the potential risk of drowning. Furthermore, we want to start a narrative on possible interventions. From this report and previous studies, it seems clear that we need to make sure that patients have not lost their ability to swim after DBS-implantation. Should we take all our dystonia patients to the swimming pool before and after receiving DBS-GPi treatment?

Study funding

None.

Ethics

Approval of this study was obtained from the Medical Ethical Committee of the University Medical Center Groningen.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.parkreldis.2022.01.023.

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