

**CASE REPORT**

Purposeless Groaning in Parkinson's Disease

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Purposeless groaning has been reported in advanced progressive supranuclear palsy. We present a case of purposeless groaning occurring as a primary complaint in a patient with advanced Parkinson's disease. Purposeless groaning is thought to be a manifestation of disinhibition and perseveration due to frontal-subcortical dysfunction. Proper recognition of this phenomenon will help clinicians to avoid unnecessary investigations and treatment (e.g., prescription of opioid medications).

Key Words Parkinson's disease; groaning; moaning.

Purposeless groaning has been reported in advanced progressive supranuclear palsy (PSP), when patients have lost ambulation.¹ Recently, we reported a case of purposeless groaning in a patient with PSP when the patient was still ambulant and conversing appropriately; the groaning worsened over time with disease progression.²

Herein, we report a patient with very long-standing Parkinson's disease (PD) with purposeless groaning as a primary complaint. We were unable to find any case reports of this phenomenon in the literature, although in a case discussion (where the final diagnosis was considered to be most likely a drug-induced movement disorder), advanced PSP and PD were mentioned in the differential diagnosis of purposeless groaning.³

CASE REPORT

A 74-year-old woman with a 37-year history of PD (diagnosed at age 37) was examined in the outpatient clinic in July 2017. Her caregiver of 22 years complained that the patient's purposeless groaning had become increasingly intrusive, occurring throughout the day, with the patient becoming quiet only upon falling asleep. This was first noticed in 2013 and then resolved, recurred in 2015, and became worse in 2017.

The patient has been regularly followed up in the clinic by a

neurologist with specialization in movement disorders (SYL) since 2009. At that time, her Unified PD Rating Scale (UPDRS) motor score was 48 (indicating severe parkinsonism). Her speech was hypophonic, slurred, and stuttering. She exhibited a moderate head tilt to the right. She needed assistance to get up from her chair and was able to walk only a few steps with close supervision. She scored 24/30 on the Montreal Cognitive Assessment (MoCA) in 2010. In 2000, the patient had undergone staged bilateral pallidotomy in China to treat dyskinesias. The procedure was complicated by speech deterioration. Her family history is remarkable as her sister (the patient's only sibling), also has PD, with a fairly "typical" course (diagnosed at age 58, and successfully treated with bilateral deep brain stimulation of the subthalamic nucleus for disabling OFF; at the time of writing, this sister is still doing fairly well, albeit with hypophonic and slurred speech, 13 years after her PD diagnosis).

Over time, there has been a gradual deterioration in the patient's condition. When seen in July 2017, there had been no recent change in her medications, which consisted of 2 tablets (100/25 mg) of levodopa-carbidopa 3 times daily, 2 capsules (100/25 mg) of controlled release levodopa-benserazide at night, 10 mg of baclofen 3 times daily, 2 mg of clonazepam at night, and 10 mg of bisacodyl at night. She had never been treated with neuroleptic medication (which can potentially cause

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tardive akathisia).³

Please see the Supplementary Video 1 (in the online-only Data Supplement) and video legend for a detailed description of the patient's purposeless groaning and examination findings. The subject has given written informed consent for the publication of this case report and the video data.

DISCUSSION

Purposeless groaning has been anecdotally mentioned to occur in advanced PD,³ although to our knowledge, there has been no detailed report of this phenomenon in the literature. We believe that the present case study will be helpful to readers encountering this phenomenon in their clinical practice and that proper recognition will help clinicians to avoid unnecessary investigations and treatment (e.g., prescription of opioid medications), as highlighted previously by Stamelou and others.^{1,2}

Purposeless groaning is thought to be a manifestation of disinhibition and perseveration due to frontal-subcortical dysfunction.^{1,2} Our patient also demonstrated the "applause sign" and a grasp response, which are additional features pointing to frontal lobe dysfunction.⁴ Studies of frontal-subcortical dementias (of which PD is a "classic" cause) suggest that the frontal signs observed in these patients result from a disruption of the extensive connections of the frontal cortex with the basal ganglia, as well as other subcortical brain structures.⁵ However, we acknowledge that although the patient demonstrated some symptoms suggesting disinhibition, the correlation between frontal cortical dysfunction and groaning remains speculative, and further study of this issue is warranted.

We speculate that disease progression likely primarily accounts for the patient's condition, with PD neuropathology involving progressively more widespread subcortical and cortical brain regions. However, it remains unclear why the phenomenon of

purposeless groaning is not more common in clinical practice, despite dementia being highly prevalent in advanced PD. We cannot exclude a contributing role of the previous pallidotomy, but we consider it unlikely that this procedure was a major contributor to the groaning since this procedure was performed 13 years prior to the onset of the groaning and since the patient's MoCA score was still in the normal range when tested ten years postoperatively.

Supplementary Video Legends

Video 1. The patient is wheelchair bound. She exhibits severe laterocollis to the right. She is able to say her name and obey simple commands (e.g., lift her arms or close her eyes). The patient is unable to complete more challenging tests of frontal lobe function (e.g., go-no go test and Luria sequence; not shown). The patient makes constant groaning sounds throughout the consultation, even when distracted, such as when undergoing the test for the "applause sign" or the test for voluntary eye movements. She seems unable to suppress the groaning when requested to do so. When asked if she feels pain, she shakes her head to indicate "no." The applause sign is positive. The grasp response is present bilaterally, but other primitive (pollicomental, palmomental, snout and rooting) reflexes are absent. She exhibits some apraxia of eye opening. Vertical eye movements are intact (not shown).

Supplementary Materials

The online-only Data Supplement is available with this article at <https://doi.org/10.14802/jmd.18004>.

Conflicts of Interest

The authors have no financial conflicts of interest.

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