Hemiparkinsonism caused by a lateral sphenoid wing meningioma, with tractography analysis: illustrative case

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BACKGROUND The etiologies of parkinsonism are diverse. A possible and rare cause of hemiparkinsonism is mechanical compression of the basal ganglia and its connecting white matter tracts. The authors present a case of hemiparkinsonism caused by a lateral sphenoid wing meningioma, discuss the underlying pathophysiology based on tractography, and systematically review the existing literature.

OBSERVATIONS A 59-year-old female was referred for a left-sided tremor of the hand, accompanied by a cogwheel rigidity of the left arm. Symptomatology appeared 1 year earlier and worsened in the previous 6 months, finally also showing involvement of the left leg. Magnetic resonance imaging (MRI) showed a space-occupying suspected meningioma originating from the right lateral sphenoid wing and compressing the ipsilateral striatum. Tractography studies contributed to elucidate the underlying pathophysiology. Resection of the meningioma could be performed without complications. At the 4-month follow-up, the patient’s hemiparkinsonism had completely recovered.

LESSONS An intracranial space-occupying lesion may be a rare cause of hemiparkinsonism. In new-onset parkinsonism, especially if a secondary form is suspected, brain MRI should be performed promptly to avoid misdiagnosis and treatment. Tractography studies help understand the underlying pathophysiology. After surgical decompression of the affected structures, symptoms can recover completely.

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KEYWORDS parkinsonism; hemiparkinsonism; meningioma; neurosurgery; fiber tractography

The most common cause of parkinsonism is Parkinson’s disease, a neurological disorder characterized by degeneration of dopamine neurons in the pars compacta of the substantia nigra (SNpc).1 However, parkinsonism is not specific to idiopathic loss of dopamine neurons and may have different pathophysiological etiologies.2-6 One possible and rare cause of hemiparkinsonism is direct mechanical compression of the basal ganglia.3-6 Different space-occupying lesions causing parkinsonism have been described in the literature: tumors, large arachnoid cysts, giant aneurysms, and chronic subdural hematoma.3-6 Here, we present a rare case of a large lateral sphenoid wing meningioma causing hemiparkinsonism. Meningiomas are relatively common intracranial tumors, comprising 36.6% of all primary central nervous system (CNS) tumors with an overall incidence of 8.3 per 100,000 persons in the Western population.7 The presenting symptoms of meningiomas, like other CNS tumors, depend upon their size and location. They are typically slow growing, and thus have an insidious symptom onset. When symptomatic, meningiomas typically present with headache, focal neurological deficits, or seizures.7 Personality changes or confusion can be seen in frontal or anterior parasagittal meningiomas, and might be initially misdiagnosed as depression or dementia.7 Parkinsonism as a presenting symptom of a space-occupying meningioma is very rare, and the underlying pathophysiological mechanisms are poorly understood. The sole compression of the basal ganglia is a too-

ABBREVIATIONS 3D = three-dimensional; CNS = central nervous system; DTI = diffusion tensor imaging; fMRI = functional magnetic resonance imaging; GPi = globus pallidus internus; JBI = Joanna Briggs Institute; MRI = magnetic resonance imaging; PRISMA = Preferred Reporting Items for Systematic Reviews and Meta-Analyses; ROI = region of interest; SNpc = substantia nigra pars compacta; SNpr = substantia nigra pars reticularis; STN = subthalamic nucleus; WHO = World Health Organization.

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patient improved slightly. At the planned follow-up 4 months after surgery, the patient exhibited no new neurological de-

sion, the more advanced understanding of the underlying pathophysiology becomes possible. The aim of this report is to present a case of tumor-induced hemiparkinsonism, to discuss the pathophysiological mechanisms, and finally to put our observations into context within the existing literature, which was systematically reviewed.

Illustrative Case

A 59-year-old female was referred by her neurologist with a left-sided tremor of the hand accompanied by fine motor skills impairment. The symptomatology had been present for 1 year and rapidly worsened in the last 6 months. Recently she also noticed a slight tremor of her left leg. The patient had no headaches or signs of increased intracranial pressure. On clinical examination, a resting and intention tremor of the left hand as well as a cogwheel rigidity with bradykinesia of the left arm was observed. A moderate and intermittent tremor of the left leg was also noticed, however without rigidity. Pyramidal signs were absent. MRI showed a large extra-axial mass on the right frontal side (4.4 × 4.6 × 4.7 cm), arising from the lateral sphenoid wing and causing a 4-mm midline-shift to the left. The striatum appeared to be the main structure compressed by the meningioma (Fig. 1A and B). To reconstruct white matter fiber tracts, DTI sequences were analyzed using the Elements Fiber-tracking version 2.0 (Brainlab GmbH). DTI sequences (28 slices, acquisition matrix 142 × 142, voxel size: 1.8 × 1.8 × 4.0 mm³, slice thickness 4.0 mm, repetition time 3,000 msec, echo time 94 msec, B = 1,000 mm²/sec, 20 directions) were obtained with the 3T Skyra MRI (Siemens). In addition, an isotropic 1-mm³ T1-weighted three-dimensional (3D) volume was acquired for subsequent definition of anatomical regions of interest (ROIs) and visualization of overlaid fiber tracts. Only fiber tracts connecting to two or more ROIs were overlaid on the anatomical image. Analyzing the relevant surrounding white matter tracts, we found that the anterior corticostriatal tracts (Fig. 2) and anterior thalamocortical radiations (Fig. 3) on the right side were strongly compressed and distorted by the suspected meningioma. The right nigrostriatal tract was not directly compressed, but rather slightly displaced posteriorly (Fig. 4). No relevant compression or displacement of the corticospinal tract was observed (Supplemental Fig. 1). Microunsurgical resection of the tumor through a right-sided pterional approach under neuromonitoring (motor and somatosensory evoked potentials) could be performed without complications achieving a Simpson grade I resection. Intraoperatively drawn tumor tissue showed a World Health Organization (WHO) grade I meningioma (methylation-class “benign-2”). Postoperatively the patient exhibited no new neurological deficits, and the left-handed tremor improved slightly. At the planned follow-up 4 months after surgery, the patient’s hemiparkinsonism had recovered completely. Postoperative MRI showed a complete tumor resection (Fig. 1C and D).

Discussion

Literature Review

The systematic literature review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines8 and adhered to a population, intervention, comparison, and outcome protocol.9 The PubMed and Embase databases were searched for relevant publications up to the end of February 2022. For the database search, a combination of the following keywords was used: “meningioma,” “parkinsonism,” “hemiparkinsonism,” and “extrapyramidal.” All surgical case reports and case series reporting parkinsonism as a presenting symptom of meningioma were included. Studies reporting lesions other than meningiomas were excluded. Only full-text articles in English were considered. Qualitative analysis of the identified publications was performed using the Joanna Briggs Institute (JBI) critical appraisal tools for use in systematic reviews (checklist for case reports).10 Publications were included in the literature review if appraised of sufficient quality according to the JBI
checklist. Outcomes are provided as percentage of cases with reported data for each outcome parameter.

Of 183 identified publications, 33 met the inclusion criteria (Supplemental Table 1). The study identification and selection process is summarized in the PRISMA flowchart (Supplemental Fig. 2). Twenty-nine of the included publications (88%) were case reports, and the remaining 4 (12%) were retrospective case series. In the last 10 years, the number of publications reporting the association between parkinsonism and a space-occupying meningioma increased considerably (Supplemental Fig. 3), attesting to the increasing awareness for this rare causality. The 33 included publications correspond to a cumulative cohort of 42 patients. Mean age of the cohort was 60 years (range, 41–82) with 61% female patients. The majority of the meningiomas causing parkinsonism were large (>4 cm in diameter) and compressed the basal ganglia (88%). They arose from the fronto-parietal convexity or falx (52%), sphenoid wing (26%), anterior cranial fossa (7%), posterior fossa (7%), middle fossa (5%), and ventricle (2%).

Tremor was the most common presenting symptom (88%), followed by rigidity (79%) and bradykinesia (62%) (Supplemental Fig. 4). Hemiparesis was only present in 10% of the cases. Almost half of the patients (45%) received presurgical medical treatment for the extrapyramidal symptoms (due to misdiagnosis in 87% of the cases); however, medical therapy rarely led to an improvement of the symptomatology (partial response in only 7% of the cases). Preoperative DTI analysis was performed in only one case (2%). At some point, all patients underwent resection of the meningioma. After surgery, 62% of the patients experienced complete recovery, 35% partial recovery, and 3% didn’t recover from parkinsonism.

Observations

Hemiparkinsonism as presenting symptom of a space-occupying meningioma is a rare phenomenon. Besides meningioma, other entities such as various CNS tumors, large arachnoid cysts, giant aneurysms, cavernoma, and chronic subdural hematoma have been reported to potentially cause secondary parkinsonism. The mechanisms causing extrapyramidal symptoms can be direct pressure to the basal ganglia nuclei, compression of midbrain structures, or damage to white matter tracts connecting the basal ganglia and cortical/mesencephalic areas. In our literature search, we found only one publication reporting the use of DTI analysis with the aim to visualize the corticospinal tract for surgical planning, rather than to understand the pathophysiology. In the following, we discuss some pathophysiological mechanisms leading to this patient’s hemiparkinsonism based on our DTI analysis. In this specific case, we assume hemiparkinsonism is multifactorial and caused by compression/displacement of the following structures (Fig. 5): striatum (putamen and caudate nucleus), anterior corticostriatal tracts, claustrum, anterior limb of the internal capsule (comprising anterior thalamic radiations), and nigrostriatal tracts.

Striatum

The pressure on the striatum (Fig. 1A and B) may cause dysfunction of dopaminergic neurons. Dopamine depletion, like in Parkinson’s disease, shifts the balance of basal ganglia activity toward the indirect pathway. This leads to excessive activity of the subthalamic nucleus (STN) that overstimulates the globus pallidus internus (GPi) and the substantia nigra pars reticulata (SNpr). Excessive activity of the STN, GPi and SNpr is plausible in this clinical case because these structures are spared from the tumor’s compression (Fig. 1A and B). Increased output from the GPi/SNpr over-inhibits the thalamocortical projections (which are already compressed at the level of the anterior limb of the internal capsule, Fig. 3), reducing cortical neuronal activation associated with movement initiation.15–17
Corticostriatal Tracts

Corticostriatal connections carry information from a variety of cortical and subcortical regions and terminate in the striatum. They play an important role in the development of smoothly executed goal-directed behaviors, including the motivation and cognition that shape the final motor tasks. The cortex projects topographically to the striatum. Thus, different striatal regions are associated with these different functions: the ventral striatum with reward, the caudate nucleus with cognition, and the putamen with motor control. In their study analyzing the organizational patterns of the corticostriatal system, Verstynen et al. demonstrated that fibers originating in more ventral areas of the central region (i.e., upper body regions) terminated in more anterior areas of the striatum compared to fibers that originated from dorsal areas of the central region (i.e., lower body). Furthermore, Staempfl et al. demonstrated in an fMRI study a certain somatotopic organization of the putamen. Hand and face activation was observed in voxels anterior to those engaged during foot actions. The hemiparkinsonism symptoms of our patient were predominantly present in her left upper extremity, which can be explained by the somatotopic organization of the corticostriatal system and the more anterior compression of corticostriatal tracts by the meningioma (Fig. 2).

Claustrum

Joutsa et al. studied cases of new-onset parkinsonism caused by focal brain lesions (located in a variety of different cortical and subcortical locations). When analyzing the connectivity of these cases, lesions causing parkinsonism were connected to several regions, but the most sensitive and specific connectivity was to the claustrum. These findings highlight the claustrum as a potential key region in the development of parkinsonism. When analyzing the MRI of our patient, the claustrum cannot be identified due to the massive tumor compression. We assume the claustrum compression to be a relevant and early cofactor in the development of our patient’s hemiparkinsonism.

Anterior Limb of Internal Capsule

The anterior limb of the internal capsule comprises mainly anterior thalamic radiations. These thalamocortical tracts include bidirectional projections between the medial frontal/orbitofrontal gyri and the anterior and medial thalamic nuclei, all of which are components of the limbic system. Thus, the anterior limb of the internal capsule participates in emotional and affective regulation, cognition, and executive function. Furthermore, it is known that thalamocortical radiations that extend to the motor cortex are implicated in movement disorders such as Parkinson’s disease. In this clinical case, we observe a relevant compression of the anterior limb of the internal capsule, thus to thalamocortical tracts (Fig. 3), being an important contributor to our patient’s contralateral hemiparkinsonism.

Nigrostriatal Tract

The nigrostriatal tract is a dopaminergic pathway, that connects the SNpc with the dorsal striatum. The nigrostriatal tract is critical in modulating movement as part of the motor loop of the basal ganglia. Compression of the nigrostriatal tract and subsequent decreased dopaminergic input to the respective striatum can lead to parkinsonism. In our case, the right nigrostriatal tract was not directly compressed, but rather slightly displaced posteriorly (Fig. 4), which could be a cofactor in the development of our patient’s symptomatology.

Strengths and Limitations

To the best of our knowledge, this is the first report of hemiparkinsonism caused by a space-occupying meningioma with a comprehensive DTI analysis that clarifies its pathophysiology. This illustrative case aimed to increase the awareness of this rare association and to emphasize the utility of tractography. The main limitation of this study lies in its nature. A case report cannot provide strong evidence for the hypothesized causative pathophysiology described here. Furthermore, the significance of the systematic literature review presented remains limited and relies mainly on the quality of the included case reports/series.

Lessons

Hemiparkinsonism as presenting symptom of a space-occupying intracranial lesion is a rare and not well-known phenomenon. In new-onset parkinsonism, especially if an atypical or secondary form is suspected, a brain MRI should be performed promptly to avoid misdiagnosis and treatment. Tractography studies can help understand the causative pathophysiology. After resection of the space-occupying lesion, symptoms can recover completely.

References


**Disclosures**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**Author Contributions**

Conception and design: Rychen, Saemann, Taub, Mariani, Soleman. Acquisition of data: Saemann, Busch, Westermann, Granzier. Analysis and interpretation of data: Rychen, Saemann, Busch, Westermann, Granzier. Drafting the article: Rychen, Saemann. Critically revising the article: Rychen, Busch, Taub, Granzier, Guzman, Soleman. Reviewed submitted version of manuscript: Rychen, Saemann, Busch, Guzman, Soleman. Approved the final version of the manuscript on behalf of all authors: Rychen. Statistical analysis: Saemann. Administrative/technical/material support: Mariani. Study supervision: Guzman, Mariani, Soleman. Sponsor of the study: Mariani.

**Supplemental Information**

Online-Only Content

Supplemental material is available with the online version of the article. [Supplemental Table and Figures.](https://thejns.org/doi/suppl/10.3171/ CASE22398)

**Previous Presentations**

Part of this study was presented as an e-poster at the EANS Meeting in Belgrade, Serbia, in October 2022.

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