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Secondary parkinsonism caused by chronic subdural hematomas owing to compressed cortex and a disturbed cortico–basal ganglia–thalamocortical circuit: illustrative case

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BACKGROUND Chronic subdural hematoma (CSDH) is a commonly encountered condition in neurological and neurosurgical practice, but the presence of concomitant parkinsonism is extremely rare. Basal ganglia disturbance is a well-known underlying mechanism; however, few cases present with cerebral cortex compression as the cause of symptoms.

OBSERVATIONS A 52-year-old man was referred to the authors' hospital with a 5-week history of gait disturbance and suspected Parkinson's disease. Neurological examination revealed a mask-like face, stooped posture, left-predominant rigidity, and postural instability. The authors initiated dopamine agonist administration, and brain magnetic resonance imaging (MRI) was scheduled. One week later, MRI showed bilateral CSDHs. The hematomas markedly compressed the bilateral cerebral cortex, whereas the midbrain and basal ganglia structures were intact. The patient underwent burr hole drainage and was discharged after 9 days without sequelae.

LESSONS CSDH can cause parkinsonism by compressing the cerebral cortex, which is a part of the cortico–basal ganglia–thalamocortical circuit. Surgery leads to positive outcomes, as illustrated by this case, in which cerebral cortex compression caused parkinsonism.

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KEYWORDS chronic subdural hematoma; secondary parkinsonism; cortico–basal ganglia–thalamocortical circuit

Parkinsonism results from the loss of dopamine-containing neurons and causes movement abnormalities, including tremor, slow movement, impaired speech, and muscle stiffness. The most common cause of parkinsonism is Parkinson's disease (PD). However, there are several other causes of parkinsonism (secondary parkinsonism), including medications, head trauma, neurodegenerative disorders, exposure to toxins, brain lesions, and metabolic disorders.[1] Some of these causes are curable and require detailed interviews and qualified imaging studies for diagnosis.

Chronic subdural hematoma (CSDH) is a common neurosurgical condition often observed in the chronic phase following traumatic brain injury and is associated with headaches and cognitive impairment. However, CSDH is rarely associated with parkinsonism.[2–6] CSDH-induced parkinsonism can arise from basal ganglia or midbrain compression and can lead to a decrease in dopaminergic neurons.[7–9] Here, we present a case of parkinsonism secondary to bilateral CSDHs successfully treated after surgical hematoma removal. Interestingly, this case's hematomas significantly compressed the cerebral cortex, while the midbrain and basal ganglia structures remained unaffected. In this rare case, parkinsonism was caused by cortical compression leading to the disruption of the cortico–basal ganglia circuit.

Illustrative Case

A 52-year-old man was referred to our hospital with a 5-week history of gait disturbance and suspected PD. He had a medical history of hypertension, for which he was prescribed antihypertensive medications by his family physician. The patient reported no history of head injury. However, neurological examination revealed a mask-like face, slightly stooped posture, left-predominant rigidity, and postural instability. PD was suspected on the basis of clinical symptoms, and dopamine agonist therapy (oral administration of pramipexole hydrochloride hydrate 0.375 mg per day)

ABBREVIATIONS CSDH = chronic subdural hematoma; CT = computed tomography; MRI = magnetic resonance imaging; PD = Parkinson's disease.

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was initiated. The patient was scheduled for brain imaging 1 week later and underwent brain computed tomography (CT) and magnetic resonance imaging (MRI), which revealed bilateral CSDHs (Fig. 1A–C). A this point, the patient admitted having struggled with alcohol addiction in the past and having had head trauma 2 months before visiting the local clinic.

The patient was subsequently admitted to our hospital for burr hole drainage. Upon admission, he presented with a mask-like face, slightly stooped posture, left-predominant rigidity, and postural instability, similar to the symptoms observed during the initial examination at our hospital. Because the CSDHs were symptomatic, surgical treatment was recommended. The subdural hematomas were removed through burr hole drainage on the day of admission. The subdural drainage catheter was removed on day 2 of hospitalization, and rehabilitation was initiated. All parkinsonism symptoms—including mask-like face, stooped posture, rigidity, and postural instability—resolved immediately on the day following surgery. Dopamine agonist therapy was discontinued on day 5 of hospitalization without subsequent symptom worsening. The patient was discharged on day 9 of hospitalization without recognizable neurological disturbances, and follow-up brain CT and MRI showed no hematoma recurrence (Fig. 1D–F).

Discussion

Observations

The patient had a history of head trauma and subsequently developed PD, which improved after removing the CSDHs. Therefore, we diagnosed this case as secondary parkinsonism due to CSDH. Idiopathic PD is the commonest form of parkinsonism, and secondary parkinsonism is present in 14%–16% of all cases involving patients older than 40 years.[10] Neurologists and neurosurgeons should consider secondary parkinsonism during the differential diagnosis, although it is relatively uncommon.

In 2012, Gelabert-Gonzalez et al. reviewed secondary parkinsonism cases due to CSDH that had been reported since 1900.[5] The authors summarized 23 cases, of which 4 were first reported and 19 had been previously reported. Since then, 2 additional cases have been reported;[11,12] thus, the total number of cases reported to date is 25, including those with unilateral hematoma (n = 15) and bilateral hematomas (n = 10). These numbers suggest that secondary parkinsonism due to bilateral CSDHs is rare.

CSDH can cause parkinsonism through mechanical pressure on the basal ganglia, midbrain compression due to herniation, decreased numbers of dopaminergic neurons in the striatum due to mechanical pressure, and circulatory disturbances in the basal ganglia due to displacement and compression of the anterior choroidal artery.[7–9] Previous reports have suggested that unilateral hematomas usually lead to contralateral parkinsonism.[6,13] However, some reports have revealed unique symptoms. Roh et al. presented a case of unilateral hematoma and ipsilateral parkinsonism,[11] and Suman et al. reported a case of bilateral hematomas and symmetrical parkinsonism.[3] Upon examination by imaging, our patient showed no midbrain compression or ischemic changes in the basal ganglia, and basal ganglia structures were preserved bilaterally; however, the hematomas considerably compressed the bilateral frontal lobes. The frontal lobes are an important part of the cortico–basal ganglia–thalamocortical circuit responsible for extrapyramidal symptoms. Circulation impairment can

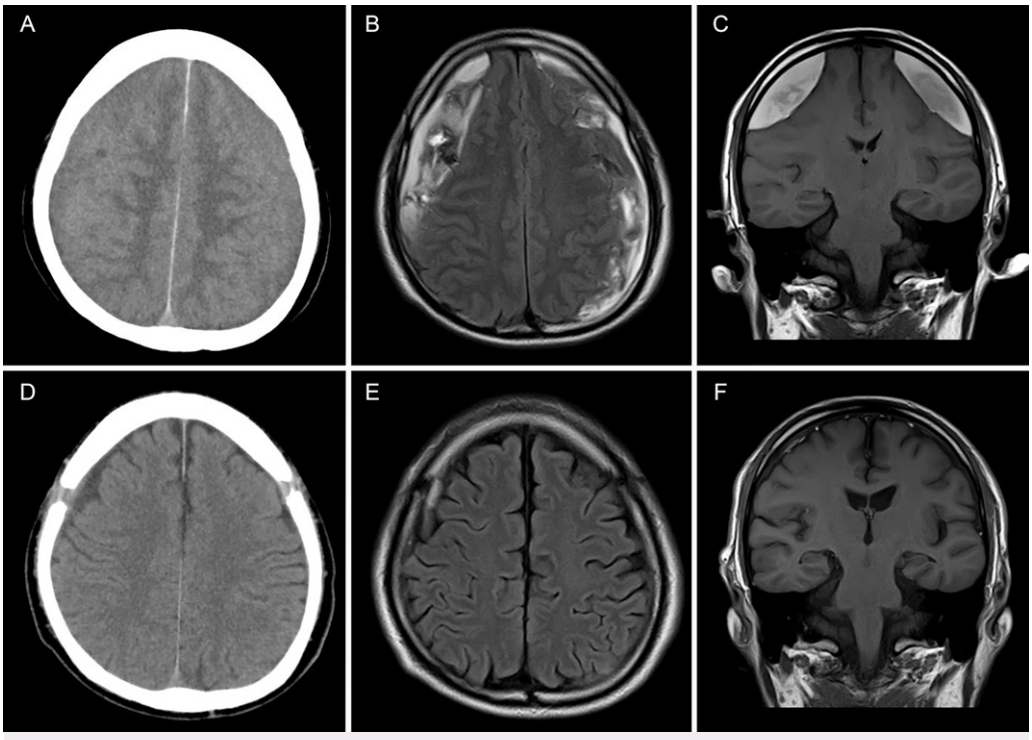

FIG. 1. Preoperative brain CT (**A**) and MRI (**B**, axial fluid-attenuated inversion recovery [FLAIR]-weighted image; **C**, coronal T1-weighted image) showing bilateral subdural hematomas compressing the bilateral frontal lobes. The compression of the midbrain and basal ganglia is negligible. Postoperative brain CT (**D**) and MRI (**E**, axial FLAIR-weighted image; **F**, coronal T1-weighted image) showing no remnant hematoma.

lead to the emergence of parkinsonian symptoms.[14] Therefore, we believe that our patient's parkinsonian symptoms emerged secondary to cortico–basal ganglia–thalamocortical circuit disruption.[15] The reason for left-predominant parkinsonism is unclear; however, the observed hematomas might have affected the right-side predominance.

Our patient showed no improvement in symptoms with preoperative drug therapy. Postoperatively, his symptoms improved rapidly, and he experienced no parkinsonism recurrence after the discontinuation of antiparkinsonian drugs. Sunada et al. reported that preoperative medication was effective in all patients with parkinsonism caused by CSDH.[13] In our patient, drug therapy might not have been effective because he had been prescribed dopamine agonist therapy (pramipexole hydrochloride hydrate 0.375 mg per day) recently, and the dose was relatively low. All previously reported cases of parkinsonism and CSDH displayed postoperative symptom improvement, similar to our patient. Therefore, surgical treatment is preferable in patients with parkinsonism caused by CSDH.

Lessons

We report a case of parkinsonism secondary to bilateral CSDHs successfully treated after surgical removal of the hematomas. CSDH can cause parkinsonism by compressing the cerebral cortex, which is a part of the cortico–basal ganglia–thalamocortical circuit. Surgery can lead to a positive outcome in parkinsonism cases caused by compression of the cerebral cortex, as illustrated by the presented case.

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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: Fukumura, Murase, Kuroda, Gon. Acquisition of data: Fukumura. Analysis and interpretation of data: Fukumura, Murase. Drafting the article: Fukumura, Murase, Gon. Critically revising the article: Fukumura, Murase, Gon. Reviewed submitted version of manuscript: Fukumura, Nakazawa. Approved the final version of the manuscript on behalf of all authors: Fukumura. Administrative/technical/material support: Nakazawa, Gon. Study supervision: Nakazawa.

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