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Case Report

Akinetic mutism following bilateral parasagittal meningioma occupied supplementary motor area removal and the spontaneous recovery of symptoms

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ABSTRACT

Background: Resection of bilateral parasagittal meningiomas of the dominant cortex is challenging. Some postoperative consequences are difficult to predict due to their low incidence. However, it is essential to recognize reversible symptoms. Akinetic mutism is a devastating but reversible symptom that occurs after supplementary motor area (SMA) injury. This report aims to provide more information to support the clinical progression of this syndrome.

Case Description: A 47-year-old woman presented with psychomotor retardation and subtle weakness, particularly on the left side. A palpable mass was identified at the head vertex. Magnetic resonance imaging revealed bilateral parasagittal meningiomas with bone and sinus invasion of the SMA. A craniotomy was performed to remove the intracapsular tumor. Two days after the operation, the patient developed gradual deterioration in her motor function until it became a lock-in-like syndrome. Then, 1.5 months after treatment in the hospital and rehabilitation unit, she gradually improved her motor, cognitive, and psychomotor skills. Total recovery was achieved after 1 year.

Conclusion: Surgery for lesions involving bilateral SMA can cause akinetic mutism. The typical manifestation of this syndrome may be devastating. However, it is reversible, and patients can regain full motor and cognitive functions over time without specific treatments. It is crucial to persevere and continue to provide the best care to the patient until recovery.

Keywords: Akinetic mutism, Craniotomy, Magnetic resonance imaging, Parasagittal meningioma, Supplementary motor area

INTRODUCTION

The removal of parasagittal meningioma poses challenges for neurosurgeons, with complication rates potentially reaching as high as 23%.[7] The involvement of the major sinuses is a concern due to the devastating consequences of venous infarction, brain edema, and hypoperfusion syndrome.[11] Bilateral parasagittal meningiomas have a far higher risk of this specific complication, as tumors often invade the superior sagittal sinus, and sinus reconstruction may be indicated.[3,9,16] Logically, the anterior 1/3 of the superior sagittal sinus can be sacrificed without significant deficits in many cases, whereas the rest cannot.[5]

The supplementary motor area (SMA), part of Broadmann area 6, is the eloquent area located on the posterior 1/3 of the superior frontal gyrus, the medial surface anterior to the premotor cortex. It is thought to be responsible for initiating movements. Some connections of the SMA to the subthalamic nucleus are presumed to be a hyperdirect pathway that suppresses thalamic circuits, resulting in movement arrest. SMA syndrome is a group of symptoms that occur after SMA function is altered.[12,13] It has various clinical presentations, such as weakness of the extremities and global akinesia, with preservation of consciousness or akinetic mutism. A remarkable characteristic of the SMA syndrome is its reversible pattern, which progresses slowly.[12,17]

Meningiomas occupying bilateral SMA have become troublesome. The risk of major venous injury and the awareness of SMA damage are critical factors. Total resection is considered impossible due to concerns regarding cortical disturbances. However, injuries are unavoidable. Due to this rare entity, many neurosurgeons are unfamiliar with its symptoms. This report describes the syndrome of akinetic mutism following bilateral parasagittal meningioma in the SMA after resection and how symptoms progress afterwards.

CASE PRESENTATION

Clinical presentation

A 47-year-old woman presented with psychomotor retardation and subtle weakness, particularly on the left side. Her speech was comprehensible but slow. No cranial nerve palsy was observed, and pupillary responses were preserved. She did not complain of headache but had an abnormal palpable, growing deformity in the frontal bone.

Diagnostic imaging

Magnetic resonance imaging (MRI) revealed a bilateral parasagittal meningioma invading the SMA. Bone hyperostosis with the signal intensity of the tumor beneath the scalp was shown. The overall extension area of the tumor involved both SMA cortices, completely occluded the middle 1/3 of the superior sagittal sinus area, and burst into the covered bone [Figure 1]. Magnetic resonance venography (MRV) was performed 1 day later to assess sinus patency. No filling signal of the sinus around the tumor area was demonstrated on MRV, indicating total sinus occlusion [Figure 2]. Mild edema was observed around the tumor margins. All laboratory test results were normal.

Operative procedure

A craniotomy was performed to achieve near-total tumor removal. Initially, the tumor was identified as invading the outer cortex of the bone and integrating with the dura mater. A doughnut-shaped craniotomy was performed. The island of bone with the midline tumor was carefully drilled and removed. The tumor completely penetrated the sagittal sinus at the point where it originated, and no sinus wall was identified in the tumor. No venous bleeding was observed from the incision. Internal debulking was also performed, eliminating all tumor masses in the area, including those within the invaded sagittal sinus. The left-only tumor plugged the sagittal sinuses anterior and posterior to the lesion, which was still intact, and a thin layer of the capsule was left in place to avoid eloquent cortex injury. A pericranium graft was used, and skin closure was performed. We did not perform cranioplasty immediately after tumor removal because we considered the patient to be at high risk of postoperative cerebral edema.

Clinical progression

The patient was conscious and could follow instructions during an immediate postoperative evaluation. Motor strength decreased in the right leg, but the patient could still raise it against gravity. Conversely, the strength of all other extremities was intact. Extubation was performed

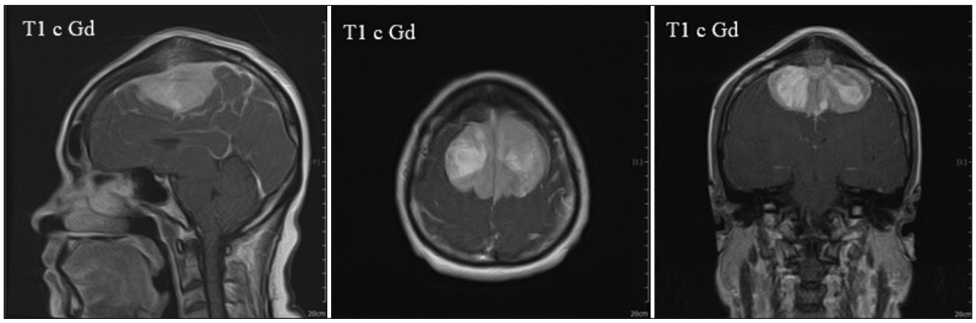

Figure 1: T1-weighted imaging with gadolinium injection revealed bilateral parasagittal meningiomas occupying both supplementary motor area areas. Bone hyperostosis was observed with a slightly increased intensity in the bone cortex, which reflected tumor infiltration.

after 24 hours of observation. However, the patient's motor response slowed several hours later, although she could still communicate. Motor function progressively deteriorated until she could not raise her extremities. The following day, the patient's motor function was completely unresponsive to external stimuli. Her eyes blinked and moved spontaneously in response to voices and commands. There were no movements of any limbs or head. The patient was reintubated to prevent airway collapse and sent for computed tomography (CT) to evaluate a possible infarction or other complications. No large infarcts are other than a moderate degree of perioperative edema were observed. A brain MRI was done a day after the patient's clinical condition became stable, and no infarction showed in diffusion-weighted imaging. A follow-up CT did not show any hemorrhage. Pathological study revealed a meningothelial meningioma (the World Health Organization grade I).

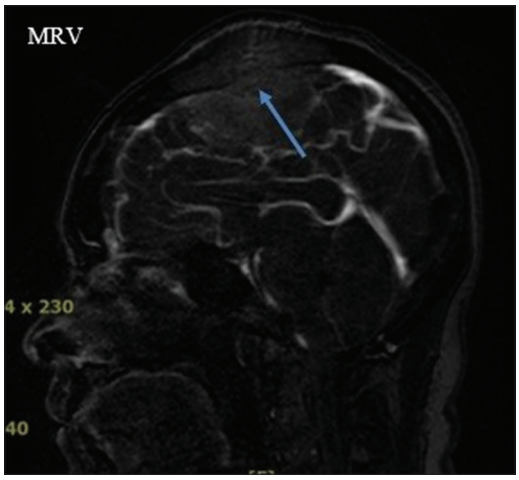

Figure 2: Magnetic resonance venography (MRV) showed no signal intensity at the middle 1/3 of the superior sagittal sinus (long arrow), indicating total occlusion.

She stayed in the intensive care unit for 1 month and had spontaneous eye-opening on voice calls or light. This process seemed static until the end of the month, when her eye responses became more frequent, and some motor twitching was observed. After 1.5 months, she was transferred back for rehabilitation at a local hospital. The first recovery was observed in the right and left legs.

The recovery process accelerated afterwards. The upper extremity function returned. In a couple of weeks, the patient progressed to ambulation, recognized more people, and began communicating faster 2.5 months after the operation. She visited a clinic and was able to walk in with slight difficulty. Her response was faster than the preoperative status. Speech fluency improved, and psychomotor retardation noticeably disappeared. After all symptoms progression and clinical improvement, the patient was diagnosed with akinetic mutism. Follow-up MRI was done at 3 months, showing minimal residual along the falx cerebri [Figure 3]. One year after the surgery, the patient fully recovered her motor, sensory, and cognitive functions.

DISCUSSION

Akinetic mutism is caused by pathologies in several areas of the brain, especially the mesencephalic-diencephalic region, cingulate gyrus, mesial frontal lobe, and, more often, the SMA.[1,2] The most common clinical features include akinesia and mutism, in which patients are aware of stimuli but cannot initiate movement or respond to them. Despite this, their consciousness remained unchanged. Some of these are considered a spectrum of SMA syndrome features. Several reports have described SMA syndromes, which include the onset of deterioration, beginning around 24–48 hours postoperatively and continuing up to 3 months or more before the state of recovery is processed.[4,8,10,14,15,18] However, few of them had akinetic mutism, which was the most severe characteristic of SMA syndrome. This was a prolonged pattern, but most patients recovered exceptionally well. Several studies indicated 1 year or more to gain a complete

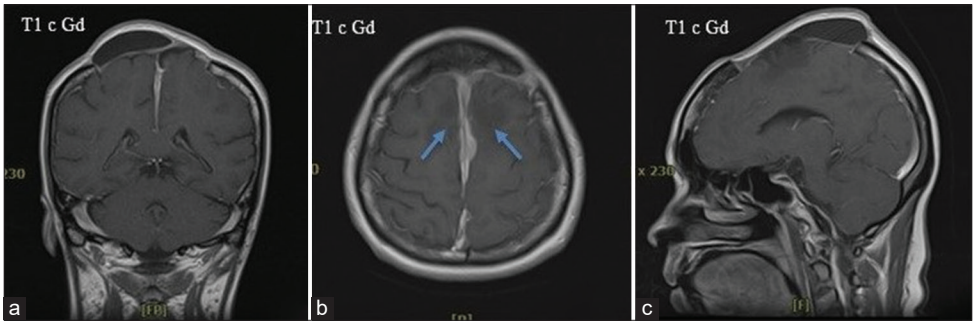

Figure 3: (a-c) Postoperative T1WI c gadolinium-enhanced MRI shows some residual tumor at falx cerebri. (b) The hypointensity area at the superior frontal gyrus around SMA (blue arrow) could cause akinetic mutism.

Table 1: Previous reports of SMA syndromes occurring after meningioma resection around SMA: The onset of symptoms, time to recover, and functional recovery of each patient.

Authors	Patient Profile	Tumor characteristics	Venous invasion	The onset of symptoms after surgery	Time to first noticeable recover	Time to gross recovery	Functional recovery
Heiferman *et al.*, 2014[8]	1. 42 y, female	Unilateral parasagittal meningioma, Right SMA	Total	2 day	1 month	1 year	Total
	2. 53 y, female	Bilateral meningioma, SMA	Total	1 week	2 week	3 month	Total
Tsai *et al.*, 2021[18]	1. 55 y, male	Bilateral	Total	0 day	7 d	7 week	Total
Berg-Johnson *et al.*, 2017[4]	1. 49 y, female	Bilateral	Total	0 day	N/A	>6 month	Partial
	2. 49 y, male	Bilateral	Total	0 day	N/A	>6 month	Partial
	3. 70 y, male	Unilateral	Partial	0 day	N/A	3 month	Total
	4. 77 y, male	Unilateral	Partial	0 day	N/A	3 month	Partial
	5. 68 y, male	Unilateral	Partial	0 day	N/A	3 month	Total
Martinez *et al.*, 2019[10]	1. 48 y, female	Bilateral	Partial	0 day	N/A	3 week	N/A
	2. 49 y, female	Unilateral	Partial	0 day	N/A	10 day	N/A
Shamov *et al.*, 2020[15]	1. 55 y, male	Unilateral	N/A	0 day	N/A	3 month	N/A
	2. 54 y, female	Unilateral	N/A	0 day	N/A	2 month	N/A
	3. 58 y, male	Unilateral	N/A	0 day	N/A	2 month	N/A
Satter *et al.*, 2017[14]	1. 65 y, female	Unilateral	N/A	2 day	1 week	6 month	Total

SMA: Supplementary motor area, N/A: Not available

recovery. This case follows the typical presentation of akinetic mutism described decades ago by neurologists.

Several authors have described the clinical presentation and progression of symptoms after surgery. Table 1 shows a literature review of cases reported worldwide. Heiferman *et al.* reported two cases where recovery began after 15 days and returned completely after 3 months.[8] Tsai *et al.* reported aphasia recovery after 7 days, while motor recovery began after 7 weeks.[18] Palmisciano *et al.*[12] reviewed milder forms of SMA syndrome after surgical resection of low-grade gliomas. Most patients develop symptoms 24 hours after surgery, with motor deficits being the most common presentation. The duration of symptoms was 45 days after onset, and 80% of patients achieved total resolution, while 20% had persistent symptoms.

The exact cause of this phenomenon can be attributed to many pathologies that affect this area. In this case, SMA injury might be caused by venous insufficiency, which, in this case, we noticed the occlusion of the main sagittal sinus from the preoperative investigation. However, the development of the surrounding cortical venous drainage system maintained adequate circulation before surgery. Another possible cause is tissue damage by mechanical forces or heat used during tumor dissection. This may have temporarily caused the eloquent area to become inflamed and edematous. After the pathology subsided, brain tissue recovered. These symptoms are reversible and motor, sensory, or cognitive.[6] Seizure was less likely in this patient since the patient remained conscious during symptoms. The pattern of deterioration was gradual, with no fluctuation of symptoms. Arterial infarction was also less likely since, in follow-up, CT and MRI did not show any signs of arterial infarction. Hypointensity in follow-up MRI 3 months after surgery accounted for the gliotic change.

There are several learning points in the present case. First, while preservation of the capsule to avoid injury to the cortex is a prudent strategy, injury still occurs due to thermal damage. Therefore, it is crucial to avoid using monopolar cautery and decrease bipolar power to the minimum effective level. Second, the cortical venous system may be essential in developing postoperative venous infarction. The collateral cortical veins drain cerebral blood circulation even when the sagittal sinus becomes occluded. Care should be taken to avoid injury to these cortical veins, particularly when the brain environment changes.

CONCLUSION

This report aimed to share data supporting the idea that akinetic mutism, which may occur after bilateral SMA meningioma resection, is a reversible phenomenon. Most patients fully recover without any neuro deficits after months. Therefore, it is critical to recognize this symptom.

Proper postoperative care should be provided to the patient regardless of consciousness level, and relevant complications, such as deep vein thrombosis or joint stiffness, should be prevented. Adjunct supportive procedures, such as tracheostomy, should be performed to avoid the risk of pneumonia, and when the clinical symptoms subside, the tracheostomy tube can be removed afterwards.

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Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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