Supplementary Motor Area Syndrome After Resection of a Dominant Hemisphere Parasagittal Meningioma: A Case Report

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BACKGROUND AND IMPORTANCE: Supplementary motor area (SMA) syndrome is a common, transient postoperative complication of intra-axial tumor resections involving the SMA and posterior cingulate gyrus. It is also reported as a rare complication of resecting extra-axial lesions. Meningiomas represent the most common, nonmalignant primary central nervous system tumor in adults, which present most commonly in parasagittal locations. Resection of dominant hemisphere parasagittal meningiomas overlying or infiltrating into the SMA region carry a recognizable risk for developing SMA syndrome postoperatively.

CLINICAL PRESENTATION: We present a 58-year-old woman with intermittent headaches and concern for new-onset seizures. MRI demonstrated an extra-axial mass involving the left frontal convexity and SMA region with homogenous postcontrast enhancement. There was radiographic involvement of the superior sagittal sinus and inner table of the skull. Fluid-attenuated recovery signal and perilesional vasogenic edema were also noted. The imaging findings favored a parasagittal meningioma, and surgical resection was performed. Arachnoid invasion and pial infiltration of the tumor over the SMA were evident during the operation.

CONCLUSION: A detailed understanding of the functional neuroanatomy and clinical pathophysiology of eloquent cortical regions is important for preoperative planning and patient counseling. Surgical resection of lesions in such areas can result in rare complications uniquely implicated in specific patient subsets. Recognizing these patients in the preoperative setting is imperative for proper counseling of patients and families.

KEY WORDS: Complications, Extra-axial lesion, Meningioma, Supplementary motor area, SMA syndrome

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Meningiomas are the most common, nonmalignant primary central nervous system tumors in adults, which present most frequently in parasagittal locations.[1-4] Surgical resection with the dural attachment is the treatment of choice for symptomatic or enlarging meningiomas, and this also provides a pathological specimen important for predicting prognosis.[1,5-9] Postoperative complication rates with parasagittal meningioma resections are reported at 15%–30%.[5,10] A rare complication is supplementary motor area (SMA) syndrome.[11]

The SMA is an area of known neurosurgical importance because it is frequently invaded by intra-axial tumors, most notably gliomas.[12,13] Surgical resection of these tumors can result in SMA syndrome.[12-15] The reported incidence of SMA syndrome reaches 60.7% with glioma resections involving the SMA, but the overall incidence with all tumor types is variable (23%-100%).[12,14]

SMA syndrome is clinically recognized as a transient contralateral akinesia or hemiparesis with preserved muscle tone developing within 24 hours after an inciting injury.[12-14,16] Speech disturbance also develops if the language-dominant hemisphere is involved and represents a motor apraxia.[12-14,17] Recovery occurs in a characteristic pattern: speech recovers first followed by the lower extremity and then the upper extremity.[13,14,16,17] SMA syndrome has a favorable prognosis with 90% of patients making a full recovery within 1 week to 6 months.[11,12,18]

We present a case of postoperative SMA syndrome after resection of a dominant hemisphere parasagittal meningioma with brain invasion.

ABBREVIATIONS: MRC, Medical Research Council; **SMA,** supplementary motor area; **WHO,** World Health Organization.

CLINICAL PRESENTATION

A 58-year-old woman presented for evaluation of intermittent headaches and concern for new-onset seizures with a nonfocal neurologic examination. Noncontrast computed tomography scan of the head demonstrated a left hyperdense extra-axial lesion concerning for a parasagittal meningioma. MRI of the brain confirmed the findings (Figure 1). The lesion was overlying the left supplementary motor area, premotor cortex, and precentral gyrus with homogenous postcontrast enhancement. There was involvement of the superior sagittal sinus and inner table of the skull. There were also fluid-attenuated recovery signal and perilesional vasogenic edema. Surgical resection was pursued with the patient's informed consent. Given the lesion's location, postoperative SMA syndrome was anticipated for which the patient and family were counseled before the operation.

The patient was positioned supine and secured using the Mayfield cranial stabilization system. A paramedian horseshoe skin incision and a frontoparietal bone flap were made with neuroimaging guidance. The inner table of the bone was shaved until a tumor-free surface was obtained. The dura was opened and divided with tumor involvement present on the lateral leaflet of the superior sagittal sinus. Intratumoral debulking was accomplished. Arachnoid invasion was noted. Subsequently, the tumor was found to be infiltrating the pia and was gently removed from the invaded surface. Additional removal of tumor to decompress the midline structures was performed. The dural attachment to the superior sagittal sinus was coagulated. A pericranial flap was attached to the clean dural edge. The bone flap was securely reattached, and the incision was closed in the usual fashion. Final pathological diagnosis was a World Health Organization (WHO) Grade II Atypical Meningioma with a Ki-67 proliferative index of 15%.

Immediately postoperatively, right-sided hemiparesis (Medical Research Council [MRC] Scale for Muscle Strength grade 0) and global aphasia were present. SMA syndrome was suspected. The postoperative MRI showed gross total resection of the tumor with residual vasogenic edema and a focal cortical rim of diffusion restriction where pial infiltration was identified intraoperatively (Figure 2). The patient was managed conservatively with high-dose dexamethasone.

By postoperative day 2, the aphasia resolved, motor function in the right lower extremity began to improve (MRC Scale for Muscle Strength grade 3), and the right upper extremity remained hemiparetic (MRC Scale for Muscle Strength grade 0). By postoperative day 4, all right-sided motor functions achieved MRC Scale for Muscle Strength grade 4. By 2 weeks postoperative, the only neurological deficit was the stable, right-sided apraxia.

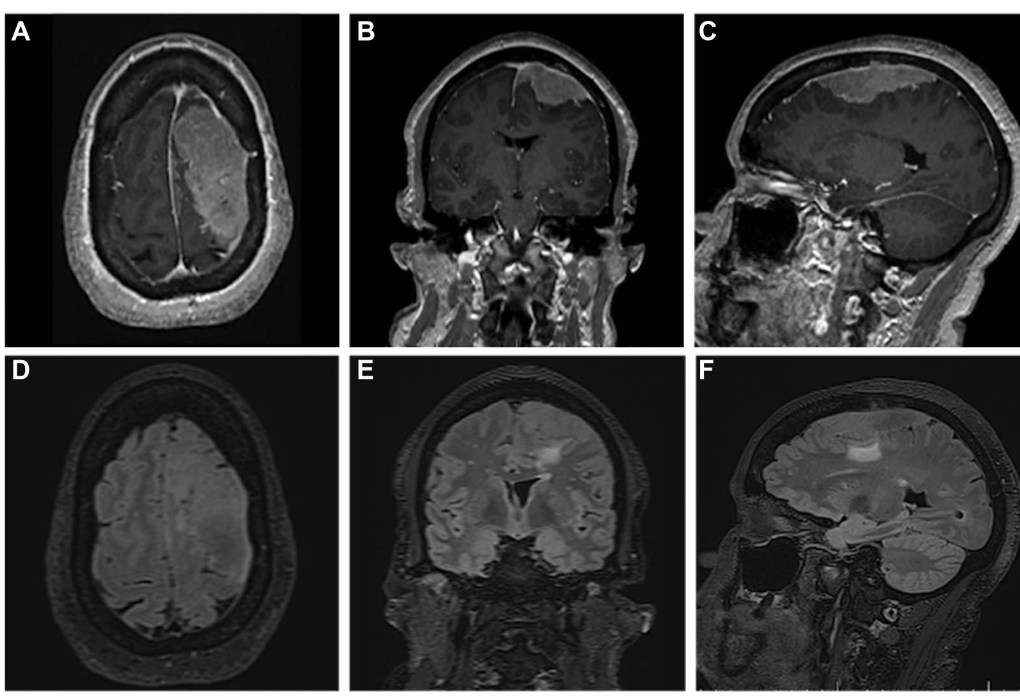

FIGURE 1. **A–F**, *Preoperative MRI sequences demonstrating a left-sided parasagittal meningioma.* **A, D**, *Axial* **B, E**, *coronal, and* **C, F** *parasagittal MRI in* **A–C**, *T1-weighted gadolinium contrast-enhanced sequences and* **D–F**, *T2-weighted fluid-attenuated recovery sequences reveal an extra-axial mass over the left frontal lobe. The extra-axial lesion measures 7.3 cm anteriorly to posteriorly, by 3.2 cm transversely, and by 2.1 cm cranial to caudal with a lobulated parenchymal margin and vasogenic edema.*

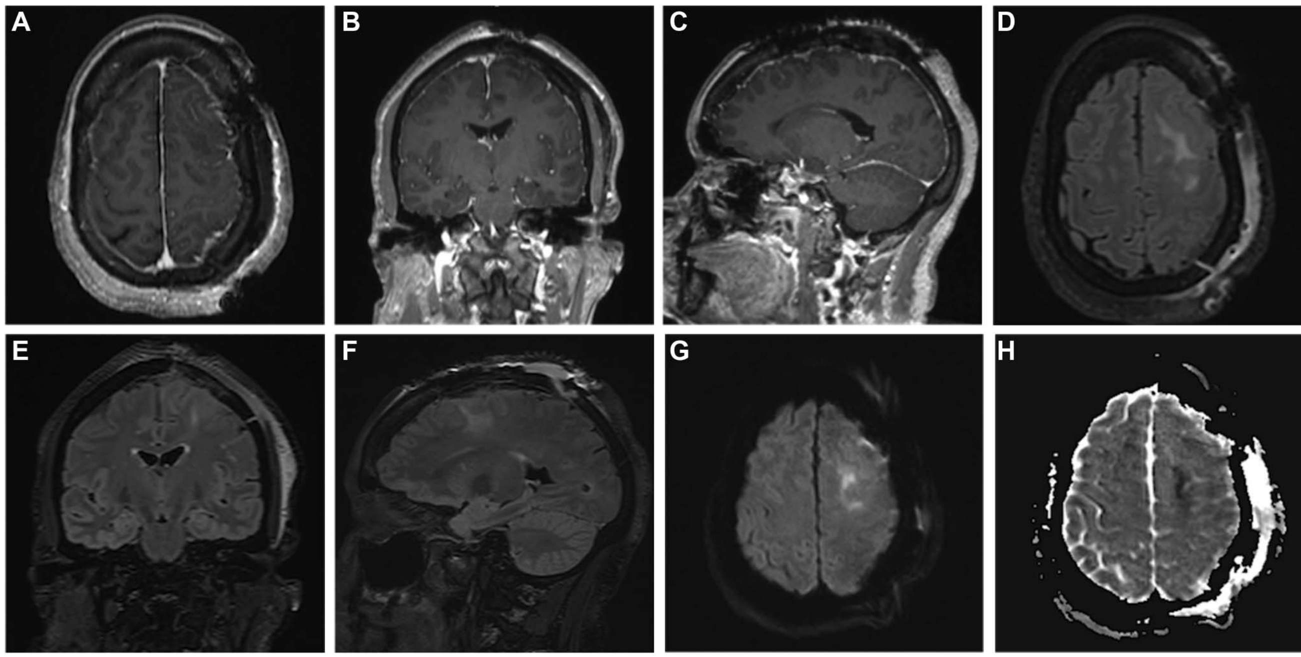

FIGURE 2. *Postoperative A–C, MRI T1-weighted gadolinium contrast-enhanced sequences and D–F, T2-weighted FLAIR sequences demonstrating gross total resection of left parasagittal meningioma. D-F, FLAIR sequences demonstrate stable to improved vasogenic edemas postoperatively. G, H, Postoperative MRI diffusion-weighted sequences show a small region of diffusion restriction over the left supplementary motor area. FLAIR, fluid-attenuated recovery.*

DISCUSSION

The SMA constitutes an eloquent region of cortex characterized by rich interconnectivity with other cortical and subcortical structures to mediate domain-general sequence processing of motor, language, and cognitive functions.[12,18-21] The anatomic boundaries of the SMA are the superior frontal sulcus (lateral), the interhemispheric fissure (medial), the cingulate sulcus (inferior), the precentral sulcus (posterior), and 5 centimeters anterior to the precentral sulcus (anterior).[12,14,22] Somatotopic organization of the SMA has a rostro-caudal arrangement.[23] The most rostral area controls the movement of the contralateral lower extremity, the middle portion controls the movement of the contralateral upper extremities, and more caudal areas involve movements of the face.[23] Language is controlled by the most caudal area of the SMA.[23] Functional division of the SMA is also conceptualized as a caudal SMA proper, which projects into the corticospinal tract, and a rostral pre-SMA, which receives input from the prefrontal cortex and the cingulate motor area.[12,14,18,19] The SMA is also an important contributor to speech and language through subcortical connections by U-shaped association fibers, the frontal aslant tract, and the frontostriatal tract.[12,14,17,21] Finally, the SMA is involved in a negative motor network, which is responsible for modulation and arrest in speech and motor function.[12,17] Severity of SMA syndrome has been correlated with damage to the posterior SMA, posterior cingulate gyrus, or frontal aslant tract.[12,14,16-18,21,22]

Postoperative SMA syndrome after parasagittal meningioma resections is a rare complication for which a single series of 12 reported cases exists.[11] In this series, the motor deficits were variable in severity.[11] Aphasia only developed in patients with left hemisphere involvement, consistent with the typical laterality of language dominance.[11,12,14,17,18] The characteristic recovery pattern of speech disturbance before motor function was identified consistently in dominant hemisphere cases.[11,13,14,16,17,24] The exact mechanism remains unclear, and additional studies are needed.

Development of SMA syndrome is increased in cases when meningiomas of higher grade or brain invasion are suspected in the preoperative evaluation. Meningiomas are risk-stratified into prognostic grades for recurrence or aggressive growth rates based on the histological subtype according to the 2021 WHO classification criteria.[2,25] Greater risk of recurrence or aggressive growth is seen with increasing WHO grade, higher proliferative indices (Ki-67, MIB-1), brain invasion, and residual tumor present after surgical resection.[6-8,10,25,26] The presence of peritumoral vasogenic edema is associated with higher-grade tumors and should raise concern for recruitment of the pial blood supply and an indistinct interface between the tumor and brain.[1,5] Additional radiological findings consistent with brain invasion include a lobulated parenchymal margin and loss of the cerebrospinal fluid cleft sign.[27] Neuromonitoring with direct cortical/subcortical stimulation and motor and somatosensory evoked potentials is recommended for intra-axial tumor resections and beneficial during surgical removal of extra-axial tumors in the SMA region to help predict the postoperative course.[12]

Our patient presented with preoperative risk factors for an aggressive and invasive tumor involving the SMA. The tumor's location involved the medial, lateral, posterior, and anterior boundaries of the left SMA. Peritumoral edema was present along the inferior border of the tumor involving the cingulate gyrus and caudal SMA. Given the above risk factors, the patient was counseled about the high likelihood of SMA syndrome in the postoperative setting. Intraoperative findings of pial infiltration and arachnoid invasion suggested a higher grade or aggressive growth pattern, which was confirmed by pathological diagnosis of an atypical (WHO Grade II) meningioma. SMA syndrome was strongly suspected as the cause of the patient's postoperative neurological deficits. The appropriate imaging diagnostic studies were obtained to ensure that no other explanation was contributing to this clinical picture. The focal cortical rim of diffusion restriction in the surgical cavity (Figure 2) at the site of pial infiltration is a known pattern consistent with inflammation after subpial microdissection rather than ischemic changes.[5] As expected, the patient's speech recovered first and completely, followed by the lower and then the upper extremity with near resolution of all symptoms at 2 weeks postoperatively.

Limitations

This report provides a case-level presentation, discussion, and literature review on preoperative evaluation, decision making, and patient counseling for postoperative SMA syndrome in patients with extra-axial tumors. Further, larger studies are needed to expand and validate clinical knowledge and mechanisms of SMA syndrome specific to extra-axial tumor resections.

CONCLUSION

SMA syndrome is a rare complication after resecting extra-axial tumors involving the SMA. The importance of applying a detailed understanding of the functional neuroanatomy of eloquent cortical regions to individualized preoperative planning and anticipatory patient counseling is exemplified by this case.

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