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Recurrent supplementary motor area syndrome in relapsed parasagittal meningiomas: from the onset to the origin☆

Recurrencia del síndrome de área motora suplementaria en meningiomas parasagitales recidivados: del evento al origen

Dear Editor:

We describe the key diagnostic and pathophysiological features of supplementary motor area (SMA) syndrome and the factors associated with good prognosis through 2 case reports.

SMA syndrome appears in patients undergoing surgery in areas close to the dorsomedial frontal lobe or SMA; it is characterised by immediate, transient contralateral motor deficits and may also be associated with language impairment.[1,2]

Although the incidence of SMA syndrome is approximately 50% of all cases of tumours at this location,[1–3] and surgical treatment of parasagittal lesions is relatively frequent, SMA syndrome is extremely rare after surgery for extra-axial tumours.[1,4–6]

Illustrative case reports

Patient 1

Our first patient was a 48-year-old woman with history of parasagittal meningioma; she had undergone surgery 10 years previously and subsequently presented tumour recurrence (Fig. 1A and B). The lesion was completely resected (Fig. 1C and D). After the procedure, the patient presented global aphasia and right hemiparesis (muscle strength 2/5). After a week receiving physical rehabilitation and speech therapy, she recovered strength in the right arm (5/5) and leg (4/5). Dysphasia resolved on day 6. Three weeks after the intervention, the patient was able to walk independently and had resumed work (modified Rankin Scale score of 0); she displayed 5/5 strength in all muscle groups.

Patient 2

Our second patient was a 49-year-old woman with history of interhemispheric meningioma, which was treated surgically 20 years previously. She underwent surgery after presenting tumour recurrence (Fig. 2A and B); the procedure was uneventful and complete tumour resection was achieved (Fig. 2C and D). Upon awakening, the patient presented normal consciousness but was unable to move the left side of her body (muscle strength 1/5). CT angiography ruled out complications of venous drainage. After a short rehabilitation period, the patient recovered baseline muscle strength (5/5); she was discharged after 10 days with no other complications.

Discussion

Surgery to the SMA may cause contralateral motor deficits and motor aphasia (the latter may appear when surgery is performed on the dominant hemisphere).[4] Unlike lesions to the pyramidal tract, SMA syndrome is transient and has good prognosis, and patients do not lose muscle tone.[6] This syndrome is distinct from the progressive, irreversible deficits that appear 24-48 hours after the procedure due to venous infarction secondary to a lesion to a parasagittal draining vein.[6]

SMA syndrome is extremely rare after surgery for parasagittal meningioma.[1,6] The pathophysiological mechanism is yet to be understood. Several hypotheses have been proposed, and include local oedema, use of retractors, microvascular damage, and resection of healthy parenchyma. Studies using functional MRI scans support the idea that the transient nature of SMA syndrome is due to neuronal plasticity.[7,8]

The cases presented here are representative in that the patients presented SMA syndrome after surgical treatment of a recurrent tumour. Abel et al.[7] reported 6 cases of intra-axial tumours located in the SMA that progressed with recurrent SMA syndrome after repeat surgery; this supports the hypothesis of neuronal reorganisation in the SMA around the previously resected areas.[4] In the case of extra-axial

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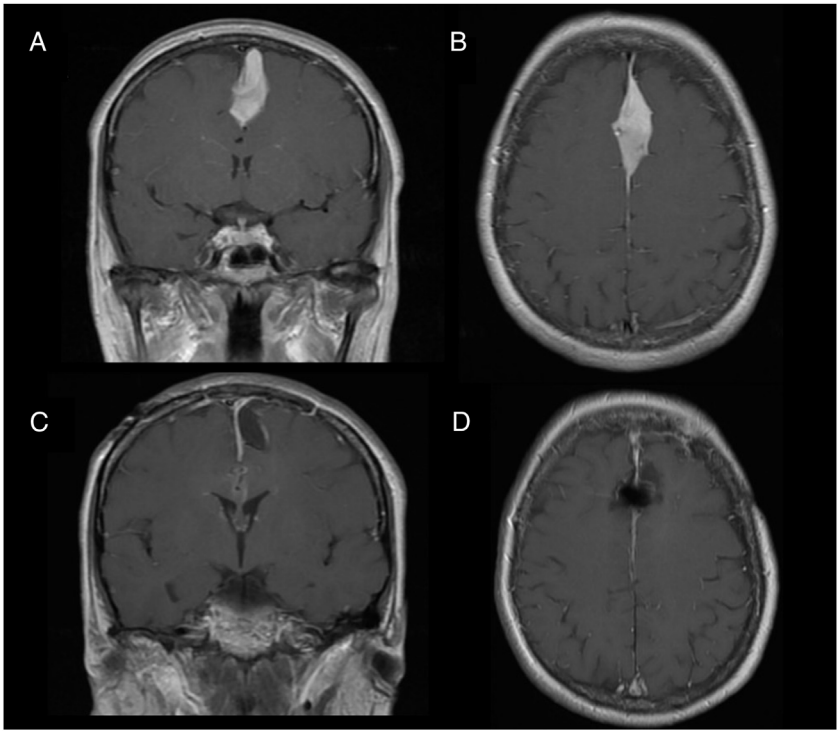

Figure 1 Patient 1: T1-weighted MRI scan with contrast administration. A and B) Preoperative images of recurrent tumour (patient recently admitted to hospital). C and D) Postoperative follow-up images.

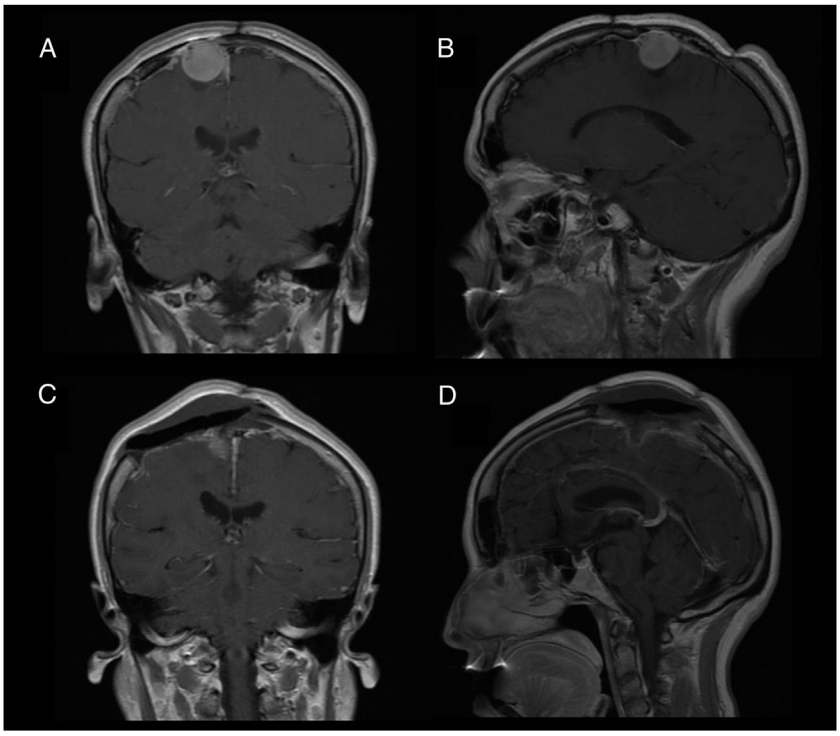

Figure 2 Patient 2: T1-weighted MRI scan with contrast administration. A and B) Preoperative images of recurrent tumour (patient recently admitted to hospital). C and D) Postoperative follow-up images.

tumours, where ideally no healthy parenchyma is resected, the hypothesis of neuronal plasticity does not seem to be plausible. We concur with Berg et al.,[6] who argue that surgical manipulation is the main cause of SMA syndrome. This is particularly relevant in cases of more aggressive tumours or tumour recurrence, where preservation of the arachnoid mater is more challenging from a technical viewpoint.[4] This suggests a certain predisposition to SMA syndrome among patients with history of surgery, who probably already present arachnoid damage, with symptoms triggered after repeat surgery.

Bibliografía

1. Berg-Johnsen J, Hogestol JA. Supplementary motor area syndrome after surgery for parasagittal meningiomas. Acta Neurochir (Wien). 2018;160:583—7. Epub 2018 Jan 23.
2. Sjober RL, Stalnacke M, Andersson M, Eriksson J. The supplementary motor area syndrome and cognitive control. Neuropsychologia. 2019;129:141—5.
3. Satter AR, Asif DS, Zannat S, Gaddam SK. Post-operative supplementary motor area syndrome: A case report. Mymensingh Med J. 2017;26(2):451—4.
4. Ibe Y, Tosaka M, Horiguchi K, Sugawara K, Miyagishima T, Hirato M. Yoshimoto Y Resection extent of the supplementary motor area and post-operative neurological deficits in glioma surgery. Br J Neurosurg. 2016;30:323—9, http://dx.doi.org/10.3109/02688697.2015.1133803. Epub 2016 Jan 13.
5. Alonso-Vanegas MA, San-Juan D, Buentello Garcia RM, Castillo-Montoya C, Senties-Madrid H, Mascher EB, et al. Long-term surgical results of supplementary motor area epilepsy surgery. J Neurosurg. 2017;127:1153—9.
6. Berg RW. Commentary: Synaptic excitation in spinal motoneurons alternates with synaptic inhibition and is balanced by outward rectification during rhythmic motor network activity. Front Neural Circuits. 2018;12:1.
7. Abel TJ, Buckley RT, Morton RP, Gabikian P, Silvergeld DL. Recurrent Supplementary Motor Area Syndrome Following Repeat Brain Tumor Resection Involving Supplementary Motor Cortex. Neurosurgery. 2015;11 Suppl 3:447—55, discussion 456.
8. Acioly MA, Cunha AM, Parise M, Rodrigues E, Tovar-Moll F. Recruitment of contralateral supplementary motor area in functional recovery following medial frontal lobe surgery: An fMRI Case Study. J Neurol Surg A Cent Eur Neurosurg. 2015;76:508—12.

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Epilepsy: a story of voices and ghosts☆

Epilepsia: una historia de voces y fantasmas

Dear Editor:

We present the case of a 51-year-old male patient, a smoker, with history of arterial hypertension and diabetes mellitus and no personal or family history of psychiatric disorders, who was brought to the emergency department after he reported hearing voices and feeling a nearby presence, which he could not see but was able to hear and feel. Both the voices and the felt presence were perceived from his left side. Furthermore, the patient's relatives reported a one-week history of short episodes of disconnection from his surroundings. The patient only reported severe headache. He did not complain with regard to the voices or the felt presence, which he considered to be real, and was unaware of the episodes of disconnection. The neurological examination revealed left superior homonymous quadrantanopia, left-sided visual extinction, and left-sided sensory extinction. An emergency head CT scan revealed a hypodense area in the right temporoparietal region, with no contrast uptake, coinciding with an MRI lesion suggestive of acute ischaemic stroke (Fig. 1). During hospitalisation, psychotic symptoms progressed to paranoid delusions of voices and felt presence. The patient was wary and became verbally aggressive when it was suggested that the voices and presence were not real. We prescribed symptomatic treatment with risperidone dosed at 1 mg/8 h. Given the atypical age of onset for primary psychosis, the co-presence of an acute brain lesion, and the occurrence of new episodes of disconnection, we requested an emergency EEG study, suspecting ictal psychosis. The EEG study, of 40 min' duration, detected 3 focal epileptic seizures lasting 1-2 min, starting in the right centroparietal region, as well as postictal slowing in the right centrotemporal region (Fig. 2). Interictal EEG revealed right temporal focal slowing. In terms of clinical signs during the episodes of paranoid delusion, the patient gazed to the left, stopping any activity, and uttered some out-of-context yet well-structured sentences;

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