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Case Report

Sudden Loss of Motor Evoked Response Followed by Clinical Recovery After Resection of an Intradural Dorsal Meningioma: A Unique Case of a Clinical-Neurophysiological Dissociation

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Abstract

In the neuromonitoring era, resection surgery of spinal intradural lesions is guided by Intraoperative Neurophysiological Monitoring (IONM) of Motor Evoked Potentials (MEP).

We describe a single case of a 70s-year-old patient who underwent surgery as a treatment for a voluminous dorsal intradural juxtamedullary meningioma: during surgery, we observed a sudden loss of bilateral lower limbs muscle MEPs, followed by a rapid recurrence of normal responses exclusively in the right side, but not in the left one. After awakening, the patient presented a transitory complete monoplegia of the left inferior limb for 30 minutes, and then it was resolved with rapid restoration of the neurological status.

Background

Spinal meningiomas represent the most frequent spinal tumor in the adult population [1,2]. The majority of meningiomas are benign tumors and there are no histopathological differences between intracranial and intraspinal meningiomas [3]; some aggressive subtypes of spinal meningiomas are associated with worse surgical and functional outcomes [4]. Intraoperative Neurophysiological Monitoring (IONM) reduces the risks of neurological deterioration after surgery of spinal meningioma [5], but, if the scope of monitoring in the presence of intramedullary lesions is clear and determined [6,7], the clinical efficacy of IONM application for the intradural extramedullary tumors rest undefinable. In a recent meta-analysis [8] of complication avoidance in resection of spinal meningiomas, IONM was applied in 4 of the 16 surgical series, and it is used in all patients in only 1 of them [9].

Sometimes, unfortunate and unexpected events occur despite the application of IONM; in many cases, and especially for our patient, determining an explanation for them remains a complex medical challenge.

Case presentation

History and examination

We reported a case of a 70-years old female patient who presented at our department with a history of different types of paresthesia interesting both feet: she reports burning, tingling, and stinging arising one year ago. Some months later, she noted

a progressive worsening in gait coordination, until the impossibility of keeping the upright position. She also described a bar-like sensation of chest constriction, associated with urinary urgency without incontinence.

The neurological exam showed gait ataxia, spastic paraparesis, hyperreflexia of bilateral patellar tendon and dysesthesia, predominant in the right limb, without segmental motor deficit: the neurological dysfunction was classified as McCormack grade 3 [10].

An urgent spine contrast Magnetic Resonance (MRI) was performed with evidence of a voluminous and solid intradural extramedullary lesion characterized by an important contrast enhancement and placed at the level of the fourth thoracic vertebra: imaging was suspected for a left dorsal-ventral-lateral meningioma. The tumor occupied 5/6 of the spinal canal, which determined a considerable spinal cord compression with displacing on the right side.

Operation and unexpected findings.

We applicated an Intraoperative Neurophysiological Monitoring (IONM) consisting of:

Transcranial motor evoked potentials (Tc-MEP): 5-7 stimuli evocated by electrodes applied on the surface of the head (min. 90 mA – max. 200 mA) produce a motor response in bilateral tibialis anterior and abductor hallucis muscles. Motor responses were also produced in the right abductor pollicis brevis muscle to control the effects of anesthesia and blood pressure.

Somatosensory Evoked Potentials (SSEP): Stimulations of the bilateral posterior tibial nerve are recorded by transcranial cortical responses (P40 potentials).

In general anesthesia, with the patient in ventral decubitus, a midline skin incision was made and a T3-T5 laminectomy was performed. After the dural incision, the large meningioma appeared with a soft consistency. The first step was to devascularize the tumor from the internal dural surface; the second step was to progressively reduce tumor volume from the left to the right side. Finally, the remaining part of the meningioma was separated without manipulation or traction of the spinal cord, observing an optimal dissection plan. After the complete resection of the meningioma, we observed a sudden loss of left lower limb MEPs (tibialis anterior and abductor hallucis muscles) and right anterior tibialis MEPs; the train of 5-7 stimuli up to 200 mA didn't evoke any motor responses. After some minutes of warm irrigation and 1 gram of Methylprednisolone administration, right anterior tibialis MEPs reappeared. This electrical status did not change until the end of the surgical procedure and left lower limb MEPs reappeared about 40 minutes after. During the operation, no variation of right abductor hallucis and right abductor pollicis brevis muscle MEPs and bilateral posterior tibial nerve SSEPs were recorded. The histological exam confirmed WHO grade I meningioma.

Outcome and follow-up

The neurological exam after awakening showed a left lower limb complete monoplegia persisting for about 30 minutes, followed by a rapid progressive recovery of the motor function until a faint monoparesis (strength 4/5 at the MRC Muscle Power Scale). The postoperative spine contrast MRI acquired 48 hours after the surgical procedure showed a total spinal cord re-expansion of the gross total resection of the tumor without complications; after 6 months a new spine contrast MRI demonstrated the absence of tumor recurrence, while the mild hemiparesis persisted without any variation.

Discussion

It is common knowledge that a transient or permanent postoperative motor deficit could occur in case of loss of Motor Evoked Potentials (MEPs) during Intraoperative Neurophysiological Monitoring (IONM) in the resection of intramedullary lesions [11]. The D-wave analysis permits to evaluate of the direct activation of fast-conducting fibres in the cortico-spinal tract [12]; considering the surgery of intramedullary tumor, less or more than 50% on the decrease in its amplitude revests an important predictor value of neurological damage.

D-wave analysis as a predictive and preventive element of post-operative neurological damage is not clear in patients who underwent surgery as a treatment for an intradural extramedullary lesion, as in the case of our patient: only a few authors show how the application of D-wave could assist the resection of spinal meningiomas [13,14].

Considering the important displacement of the spinal cord of our patient, we prefer to not apply D-wave analysis; instead, MEPs recording was accurate and highly predictive.

The first atypical event consisted of the sudden loss of bilateral lower limbs MEPs after resection of meningioma. No surgical procedure was in progress at the time of the spinal cord electrical stupor recording, and no traumatic manipulation of the spinal cord was provided during the surgery: the dissection plane permitted a gentle dissection between the meningioma and the medial surface of the dorsal spinal cord.

The second atypical event was the extremally rapid clinical recovery of left lower limb motor deficit after awakening. Some patients showed a transient worsening in neurological deficits after intradural extramedullary tumor resection, typically secondary to vasogenic edema or as a result of dissection, with resolution after on average 6 months [15,16]. The neurological exam after awakening showed a left lower limb complete monoplegia persisting for about 30 minutes, followed by a rapid progressive recovery of the motor function until a faint monoparesis. The postoperative spine contrast MRI acquired 48 hours after the surgical procedure showed a total spinal cord re-expansion of the gross total resection of the tumor without complications.

We sustain 2 different theories to explain these atypical events:

Reperfusion injury: The intradural juxtamedullary space, which became free as a consequence of the resection of the dorsal meningioma, permitted the re-expansion of the spinal cord in this cavity; it probably produced temporary oxidative stress as a consequence of the rapid revascularization of the compressed medullary segment. The clinical manifestation was

a temporary medullary stupor. This theory appears less plausible because benign meningioma is a slow-growing tumor, and reperfusion injury is more frequent after resection of rapidly compressing causes.

Cavitational effect: The rapid stretching of axons related to rapid revascularization, at the same as cerebral concussion [17] but in this case as the results of the re-expansion of the spinal cord, could cause an altered membrane conductivity, dysfunction in glucose metabolism, and a mitochondrial metabolic blockade [18]; this explains an energetic interruption of nervous conduction. Giza et al. [19,20] showed how the mitochondrial metabolic dysfunction, determined by calcium sequestration into mitochondria, provokes low production of ATP and a dysfunction of sodium–potassium pump, triggering a consequences cytoplasmatic molecular cascade that exacerbates problems related to oxidative stress and the cellular energetic crisis.

To the best of our knowledge, considering the note limitations of IONM, no authors has ever described a case of loss of MEPs during the resection of intradural extramedullary lesion, corresponding in a single transient neurological deficit with a rapid resolution. In these cases, our experience suggests that if the neurological deficit cannot be explained by a macroscopical surgical injury, such as spinal cord resection or traumatic manipulation, they may be transient with even an almost complete recovery.

Learning points/Take home messages

- IONM represents an indispensable guide in surgical treatment, especially in neurooncological spine surgery.

- Sudden IONM modifications without a clear explanation, are often sustained by a real clinical change.

- All events recorded by IONM show different meanings: true positive or false positive have to be interpreted considering the level and the site of the lesion, and clinical results after surgery.

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