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Case report

The use of facial nerve fasciculus motor evoked potential (MEP) as intraoperative neurophysiological monitoring modality in a child with a diffuse intrinsic pontine glioma: A case report

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ABSTRACT

Background: Intraoperative neurophysiological monitoring (IONM) has improved the diagnosis and surgical treatment of brainstem and posterior fossa tumors. Several modalities are available for IONM such as electroencephalography, brainstem mapping (BSM), cranial nerves evoked potentials), somatosensory evoked potentials (SEP), motor evoked potentials (MEP), brainstem auditory evoked potentials (BAEPs), nerve conduction, and electromyography (EMG) signals. Though motor evoked potential (MEP) and brainstem mapping are the most common IONM modalities used for surgical management of brainstem gliomas, cranial nerve potentials can also be of great help.

Case description: This article describes a 10-year-old child with diffuse intrinsic pontine glioma (DIPG) who presented with gradual progressive crossed hemiparesis. His brain images carried a range of potential differential diagnoses. Her underwent a successful brainstem biopsy via using motor evoked potential for facial nerve without injuring nearby structures.

Conclusion: Motor evoked potential of the facial nerve can be used solely for biopsy taking in cases of DIPG.

1. Background

Intraoperative neurophysiological monitoring (IONM) has improved the diagnosis and surgical treatment of brainstem and posterior fossa tumors [1]. Prior to the recent advances of IONM, surgical resection of posterior fossa tumors had been challenging, and the risk of long-term neurological dysfunction secondary to injury of eloquent structures had been high [2]. Intraoperative neurophysiological monitoring (IONM) allows monitoring the integrity of long tracts, locating the different cranial nerve nuclei, and monitoring the integrity of these nuclei and their cranial nerves. Such monitoring reduces the risk of damaging these structures during surgical manipulation [3]. Several modalities are available for IONM such as electroencephalography, brainstem mapping (BSM), cranial nerves evoked potentials), somatosensory evoked potentials (SEP), motor evoked potentials (MEP), brainstem auditory evoked potentials (BAEPs), nerve conduction, and electromyography (EMG) signals [3,4]. Motor evoked potential (MEP) and brainstem mapping are the most common IONM modalities used for surgical management of brainstem gliomas [3]. Intraoperative neurophysiological monitoring (IONM) does not only optimize surgical excision of posterior fossa tumors, but it also allows better diagnosis via accurate biopsy taking from debatable posterior fossa lesions without mentionable damage to critical neural tissue [5].

In this report, we present a child with diffuse intrinsic pontine glioma (DIPG) in which using motor evoked potential for facial nerve was successfully used for biopsy taking.

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2. Clinical anatomy of floor of the brainstem

Intraoperative neurophysiological monitoring (IOM) is a novel technique to identify and monitor important structures in posterior-fossa and brainstem surgery such as brainstem nuclei [1]. Mapping of facial nerve nucleus is carried out at the level of the floor of the fourth ventricle. This allows identification of safe-entry zones into the posterior fossa and brainstem [3]. On surgical opening of the dura and exposure if the floor of the fourth ventricle, a reference needle is inserted into the target muscles (i.e., the orbicularis oculi and orbicularis oris) and stimulating probe is used. A low intensity stimulation is initially used and can be progressively increased until a compound motor action potential (CMAP) is elicited. The probe is moved around the ependyma of the floor of the fourth ventricle with the lowest threshold that elicit a CMAP. It is well-known that the stimulation current can spread a few millimeters away from the probe thus allowing identification of the nearest point of the nuclei or the nerve root of the facial nerve (Fig. 1) [4].

3. Case presentation

A 10-year-old boy was referred to the neurosurgery clinic with a three-month history of left ptosis, dysphagia and right hemiparesis. The child's symptoms started three months prior to presentation with insidious progressive weakness over the right upper and lower extremities that was more evident distally. A couple of weeks after motor symptoms, the child started to develop ptosis on the left eye followed by dysphagia to solids. On physical examination, the child was slightly pale. Otherwise, his general examination did not reveal any striking features. Neurological examination revealed left partial oculomor palsy with pupillary affection, left trigeminal hypothesia, weak gag, and right pyramidal hemiparesis (crossed hemiparesis) MRC grade 3 to 4.

A magnetic resonance imaging (MRI) of the brain revealed a diffuse heterogenous mass extending from the left thalamus to the upper medulla. It was hypointense on T1 film, hyperintense on T2 and FLAIR films with no enhancement on T1 post gadolinium films. The mass resulted in pontine enlargement, displacement of the basilar artery, and moderate compression on the fourth ventricle (flattened floor) (Figs. 2 and 3). The patient's routine chemistry panel was unremarkable, and he was prepared for a surgical biopsy.

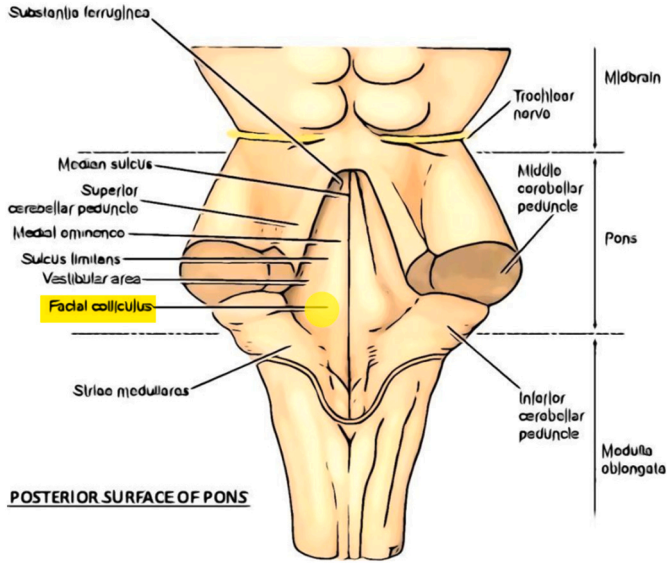

Fig. 1. Anatomical landmarks of the floor of the brainstem. Note the facial colliculus highlighted in yellow. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

For biopsy taking, a sub-occipital craniectomy was performed, and a telovelar approach was considered for reaching the floor of the brainstem. To avoid damage to eloquent structures during biopsy taking, an IONM was conducted (Fig. 4). Identification and monitoring of the MEP of the facial colliculi bilaterally was carried out throughout the surgery to avoid its injury. A surgical biopsy was, then, taken from the pontine lesion above the facial colliculi.

Histopathological examination of the biopsy revealed hyperproliferative cells with significant mitosis, nuclear atypia, and focal areas of anaplasia; a picture suggestive of grade 2 astrocytoma. Accordingly, the child was sent for radiotherapy. He was stationary postoperatively. Over the next three months, the patient received several sessions of radiotherapy. However, he developed aspiration pneumonia and died. Autopsy was not performed, because his parents refused. This case report followed the SCARE guidelines for its realization [12].

4. Discussion

The child presented is a case of diffuse posterior fossa lesion that has been challenging in both diagnosis and treatment prior to introduction of IONM. Gliomas of the brainstem are of the most common tumors among pediatric population with a peak age of onset between 7 and 9 years [6]. Focal brainstem gliomas are amenable for surgical resection, but diffuse gliomas are inoperable [3]. Even though, taking a biopsy is of considerable benefit in both focal and diffuse gliomas to identify the nature of the tumor. Brainstem gliomas consist of variable heterogenous groups of tumors, with each type having a specific growth pattern, prognosis, malignant behavior, and management plan [7]. Therefore, IONM is as important in diffuse brainstem gliomas as it is in focal potentially respectable ones [4].

Diffuse brainstem gliomas are often inoperable and associated with high risk of injury to vital neural structures during biopsy [3]. The use of IONM can help to reduce such risk [4]. In the case we present in this report, a successful complication-free biopsy was performed to a diffuse mass extending from the thalamus to the upper medulla and was sent for biopsy. In literature, several modalities have been used IONM of brainstem and/or cerebellum lesion with the MEP and brainstem mapping being the most common in cases of brainstem gliomas [3].

During the past few decades, the use of IONM became recommended in all cases of brainstem gliomas [8]. Though MRI depicts the details of tumor location, it does not help during surgery [8]. Th tumor might distort the normal anatomy of the brainstem and, therefore, obscures the known landmarks [8]. The use of IONM can overcome this fallacy by providing a continuous measurement of the function of the structures nearby the tumor to ensure their integrity throughout surgery [1,4,5].

Following the introduction and the advances in IONM during brainstem surgery, there was a notable improvement in both diagnosis and management of tumors at this narrow difficult to manipulate zone of the central nervous system [9]. Several modalities have been developed to continuously monitor brainstem neural tissue during surgery. These modalities are broadly divided into mapping, monitoring, and reflex testing [10]. In mapping, the IONM aims at identifying and differentiating different brainstem structures [10]. Monitoring aims at providing continuous assessment of the functional integrity of the neural tracts [10]. Some authors suggest the necessity of using at least two modalities for accurate IONM of brainstem tumors [5,11]. In the case presented here, however, we used a single modality that worked well, and the patient was stable postoperatively without any signs or symptoms of neurological injury.

We used mapping and monitoring modalities to identify the facial nerve colliculus to avoid it. Basically, mapping is used to localize neural structures, such as cranial nerve nuclei, cranial nerve fibers, and corticospinal tracts [9]. This is carried out via using a monopolar probe that identify these structures especially in the context of distorted normal anatomy [5]. Monitoring is carried out via one or more modalities that

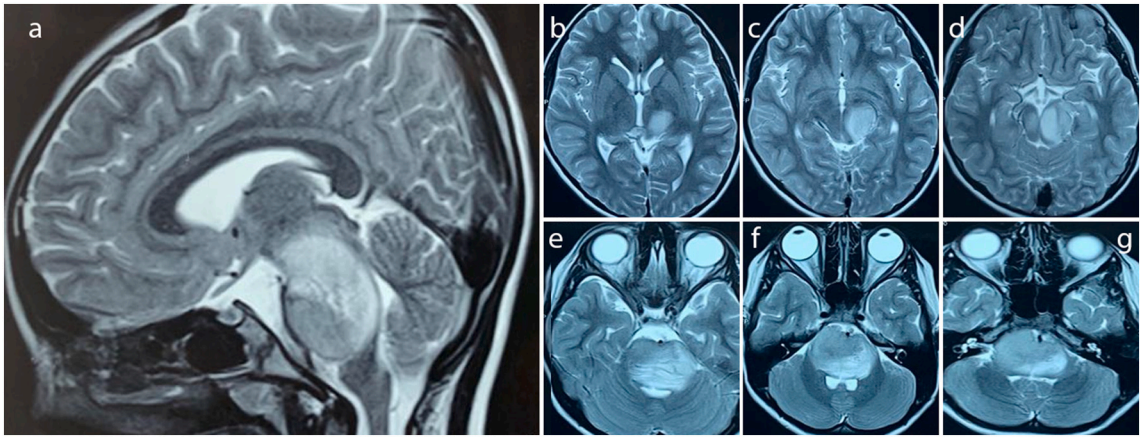

Fig. 2. MRI brain T2 sagital [a] and axial [b to g] films showing hyperintense lesion extensing from the thalamus to the upper medulla. The pons is enlarged [e, f, g], the fourth ventrile is flattened [f and g], and the basilar artery is displaced towards the left side [f and g].

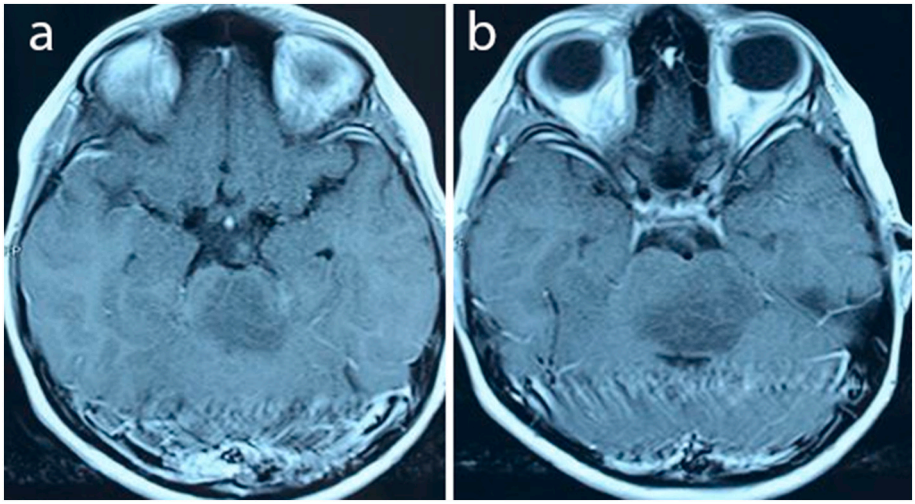

Fig. 3. MRI brain T1 post gadolinium hypointense diffuse pontine lesion showing no enhancement.

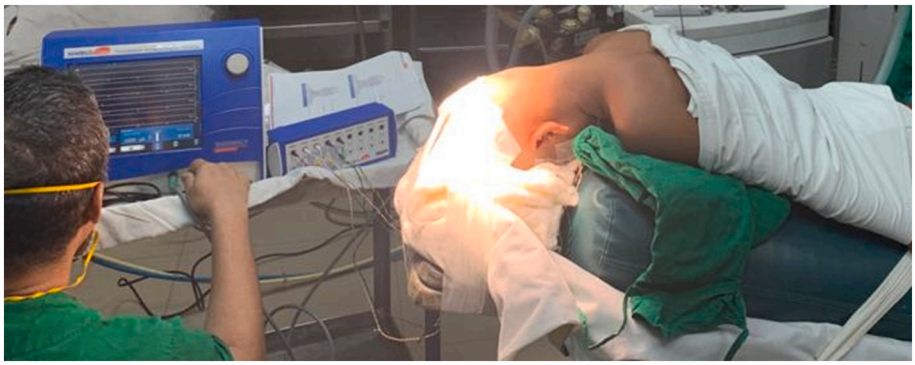

Fig. 4. Intraoperative neurophysiological monitoring (IONM) of a child with diffuse brainstem mass undergoing surgical biopsy.

continuously evaluate functional integrity of the neural structures of interest [5]. In the case presented, we used MEP to monitor the functional integrity of the facial nerve nucleus and facial fasciculus. The MEP of the facial fasciculus allowed us to ensure that the functions of the facial nerve were spared throughout surgery and the nerve was avoided during biopsy taking.

Though many other modalities may be also used, we suggest that using a single modality may also be of help particularly if the main aim of surgical intervention was biopsy-taking or partial tumor resection rather than total removal. This would allow rapid, less-time consuming, and successful biopsy-taking from the tumor. Thus, it will allow better diagnosis and consequently management of such cases.

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Ethical approval

Hospital exempts ethics approval for reported cases.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors equally contributed to the analysis and writing of the manuscript.

Research registration

Not applicable.

Guarantor

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Declaration of competing interest

All authors declare no conflict of interest.

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