Case Report

Combined extradural and intradural approach to a trigeminal nerve hemangiopericytoma with cranial nerve monitoring: a technical note of a rare case

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Abstract

Hemangiopericytoma (HPC) of the trigeminal nerve is extremely rare. We present a case of a large cystic HPC of the mandibular division of the trigeminal nerve, only the third case described in the literature, with both intradural and extradural components. We describe the surgical approach, assisted by neurophysiological techniques of mapping and monitoring including blink reflex and triggered electromyography. Additionally, we report a method of monitoring of the sensory branches of the trigeminal nerve, poorly described in the literature, through peripheral and direct nerve stimulation and recording of transcranial somatosensory evoked potentials.

INTRODUCTION

Hemangiopericytoma (HPC) of the meninges is a rare tumor, classified by WHO under the category of solitary fibrous tumor of the dura. First described in 1928 [1], most of them are locally invasive, and tend to grow along venous sinuses, which often requires adjuvant radiotherapy [2]. Recurrence as well as intracranial and extra-cranial metastases are a bad prognostic sign [3]. HPCs of the cranial nerves are extremely rare and literature revealed two reported cases of HPC of the trigeminal nerve [4]. We present a case of HPC of the mandibular division of trigeminal nerve and our surgical strategy for its resection.

CASE REPORT

A 32-year-old man presented with a 4-week history of worsening headaches, and a 1-week history of nausea and vomiting associated with memory problems over a period of months. Neurological examination was unremarkable. Computed tomography (CT) scan with contrast revealed a left temporal enhancing lesion with enlargement of the foramen ovale (Fig. 1a) and an magnetic resonance imaging (MRI) scan confirmed the presence of a large lesion (43 x 45 x 54 mm) lying in the temporal fossa, with mixed cystic and solid enhancing components (Fig. 1b). After discussion at the neuro-oncology multi-disciplinary meeting, a decision was made for surgical resection of the lesion.

Figure 1. Imaging characteristics of the V3 lesion. (a) CT scan axial, sagittal and coronal views (brain and bone window) showing the lesion in the left middle fossa with enlargement of the foramen ovale (asterix). The lesion (arrow) is uniformly enhancing with a large capping cyst causing mass effect. (b) MRI scan coronal and axial views showing a mixed solid and cystic lesion (arrow) in proximity to the Meckel’s cave.
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was identified and incised (and extra-durally (finally the foramen ovale with the tumor coming into view inter-
two layers of the dura to expose first the foramen rotundum, and (the trigeminal nerve were observed from the intradural space
whole intradural component was removed. The foramen ovale
The capsule was dissected from the left temporal lobe and the
tal file.
and the tumor was removed from that area as well. The integrity
histology consistent with HPC. The foramen ovale was widened
positive responses, and therefore we preceded to incise the tumor
orbital fissure (Fig. 2d). Dissection then proceeded between the
two layers of the dura to expose first the foramen rotundum, and
finally the foramen ovale with the tumor coming into view inter-
and extra-durally (Fig. 2f–g).
Mapping the margins using monopolar stimulation revealed no
positive responses, and therefore we preceded to incise the tumor
capsule and partially debulk the lesion. Frozen section suggested
histology consistent with HPC. The foramen ovale was widened
and the tumor was removed from that area as well. The integrity
of the all divisions of the trigeminal nerve were continuously
monitored throughout via somatosensory evoked potential (SSEP)
recordings from scalp electrodes, and via direct nerve stimulation
of the sensory and motor branches, and blink reflex monitoring
(Figs 3–6). All monitoring modalities were replicable and stable
throughout tumor removal. The dura over the temporal lobe was
then opened. The tumor was identified and extensively debulked
with a ultrasonic aspirator (Sonopet®, Stryker, Michigan USA).
The capsule was dissected from the left temporal lobe and the
whole intradural component was removed. The foramen ovale and
the trigeminal nerve were observed from the intradural space
(Fig. 2g).
Neuromonitoring parameters are described in the Supplemental
file.

HISTOPATHOLOGY
Histology showed spindle-shaped cells with large nuclei and small
amount of cytoplasm, haphazardly arranged around capillaries,
along with staghorn vessels consistent with HPC. Ki67 was found
to be ~6–7% and the tumor was graded as Grade II (WHO).
Immunohistochemistry showed negative EMA and positive
CD34.

SURGICAL TECHNIQUE
The patient was positioned supine in a Mayfield clamp, with the
head turned to the right by 45°. A skin incision was performed as
outlined in Figure 2a, and a pterional craniotomy and interfascial
approach to the temporalis was completed. The lesser and greater
sphenoid wings were drilled (Fig. 2b). The meningo-orbital band
was identified and incised (Fig. 2c).
Both anterior and middle skull base were thus exposed. The
cystic component was drained first via a dural stab incision with
a navigation guided Dandy canula, draining 20 ml of serous fluid.
This maneuver allowed decompression of the bulging temporal
lobe and facilitated further exposure of the middle fossa. Fol-
lowing that, Kerrison rongeurs were used to enlarge the superior
orbital fissure (Fig. 2d). Dissection then proceeded between the
two layers of the dura to expose first the foramen rotundum, and
finally the foramen ovale with the tumor coming into view inter-
and extra-durally (Fig. 2f–g).

FOLLOW UP
The 3-month follow-up MRI scan did not reveal any residual, and a
whole body Positron Emission Tomography (PET) scan confirmed
that there were no extracranial metastases. The patient continues
to be followed up for recurrent or metastatic disease.

DISCUSSION
HPCs are rare, comprising less than 1% of all intracranial tumors. Distinguishing them from meningiomas is critical, as these
tumors have a worse prognosis, and their rates of recurrence and incidence of extracranial disease dictate a different surveil-
ance and treatment strategy, including the frequent use of adjuvant radiotherapy. Male predominance [5, 6] and younger age [7] are key
factors in differentiating from meningiomas. Complete resection is the single most important factor in reducing the incidence of recurrence and increasing survival [5, 6, 8, 9]. However, this is often challenging due to excessive bleeding and adherence
to adjacent structures and radiotherapy is often considered post-
operatively to prevent recurrence [10]. Thus, the most recent WHO classification of tumors (2021) has assigned these lesions to the
entity ‘solitary fibrous tumor of the dura’ [11].
In this report, we present a case of V3 HPC. Our literature
search revealed only two previous reports of such tumors. Tan
et al. [5] present the case of a 41-year-old male who presented with
diplopia and facial paraesthesia and was found to have a right
sided lesion measuring 4 cm maximum diameter eroding through
Meckel’s cave and petrous apex. The second case report [4] is of a

Figure 2. Surgical technique. (a) Skin incision; (b) drilling of the lesser wing of the left sphenoid bone; (c) incision of the meningo-orbital band; (d) dura
propria and perioseal dura at the superior orbital fissure; (e) dura propria and perioseal dura at the foramen rotundum; (f) tumor coming into view at
the foramen ovale; (g) visualization of the foramen ovale from intradurally after tumor debulking.

Figure 3. Electromyography of trigeminal nerve (motor). Trigger EMG of the motor branches of the trigeminal nerve with positive motor responses (mentalis muscle EMG). Nearby cranial nerves were additionally monitored including the facial nerve CN VII (orbicularis
oculi, orbicularis oris and mentalis muscle) and abducens nerve CN IV (lateral rectus muscle).
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Figure 4. Direct nerve stimulation of the sensory V2. Direct nerve stimulation of the sensory branch V2 at the foramen ovale demonstrating cortical SSEPs recorded from scalp electrodes (montages C4’-Cz’, C4’-Fz, C4’-C3’) accordingly to the international 10–20 EEG system. Average latency: 5.4 ms, amplitude: 12.5 μV.

Figure 5. Trigeminal V2 SSEP recordings. Trigeminal V2 SSEPs were continuously recorded on the scalp using the montage C4’-Fz. Average latency: 5.4 ms, amplitude: 13 μV.

33-year-old female who presented with a seizure and a preceding 3-week history of left lower limb weakness. The underlying lesion was 5 cm across on the imaging.

Surgical considerations for HPCs of trigeminal nerve are similar to those for trigeminal schwannomas [12]. The initial approach is through a pterional craniotomy, with extradural drilling of the sphenoid wing. Unroofing of foramina ovale and rotundum allows the whole of the trigeminal complex to be mobilized. The interdural approach that we used initially is well described and gives good access to the area of the Meckel’s cave [13]. However, dural opening and intra-dural debulking was necessary in our case due to the significant intra-dural component.

Figure 6. Blink reflex. Blink reflex from left side stimulation of the supraorbital branch of the trigeminal nerve was observed with EMG activity from left orbicularis oculi muscle. Both early response (R1) and late reflex activity (R2) were recorded throughout the surgical procedure.

Neuromonitoring for trigeminal nerve commonly involves monitoring MEPs from the masseter and temporalis muscles [14]. Although monitoring orbicularis oculi muscle is mainly used to check facial nerve integrity, we used the blink reflex to monitor the function of the supra-orbital branch of trigeminal nerve, acting as the afferent arm of this reflex. We used scalp SSEPs to monitor the sensory function of trigeminal nerve with reproducible results as described in literature [15, 16].

CONCLUSIONS

We describe a rare case of HPC of trigeminal nerve. A combined extradural–intradural approach was effective in debulking this tumor, and intraoperative neuromonitoring of trigeminal nerve was a helpful adjunct in safe tumor resection.
PATIENT CONSENT
The patient has consented to the submission of the case report for submission to the journal. Case Reports is written in accordance with COPE guidelines.

CONFERENCE PRESENTATION
A part of the paper was presented as a poster in British Neuro-Oncology Society (BNOS) Annual Meeting, July 2019.

CONFLICT OF INTEREST STATEMENT
None.

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SUPPLEMENTARY MATERIAL
Supplementary material is available at JSCREP Journal online.

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