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Case Report

The usefulness of indocyanine green during surgery for hypervascular posterior fossa tumors

Takahiro Shinya#, Hideki Nagamine#, Ken-ichi Sugawara, Shogo Ishiuchi

Department of Neurosurgery, University of the Ryukyus Faculty of Medicine Hospital, Okinawa, Japan

E-mail: Takahiro Shinya - shinyashinya219@yahoo.co.jp; Hideki Nagamine - h082632@med.u-ryukyu.ac.jp; Ken-ichi Sugawara - ksuga501@med.u-ryukyu.ac.jp; *Shogo Ishiuchi - ishogo@med.u-ryukyu.ac.jp

*Corresponding author

[#]Takahiro Shinya and Hideki Nagamine equally contributed to this report

Received: 21 January 18 Accepted: 02 April 18 Published: 26 April 18

Abstract

Background: Cerebral hemangioblastomas are benign tumors with abundant blood flow that occur mainly in the posterior fossa. Tumor removal *en bloc* is important in surgical treatment because of the risk of bleeding; however, it is actually rather difficult in practice. Therefore, we propose a surgical strategy for visualizing hypervascular tumors of the posterior fossa utilizing indocyanine green (ICG).

Case Description: Case 1 involved a 48-year-old male with a history of von Hippel–Lindau (VHL) disease. Magnetic resonance imaging (MRI) revealed a solid tumor measuring 3.0 cm in diameter in the right cerebellopontine angle. We performed surgery because the tumor was pressing against the brainstem. Surgery was performed via the posterior subtemporal transtentorial approach in order to visualize the feeding artery and draining vein intraoperatively. The vessels were confirmed by ICG and the tumor was removed *en bloc*. Case 2 involved a 30-year-old woman. Signs of increased intracranial pressure were noted, and an MRI revealed a solid tumor 3.5 cm in diameter in the left cerebellar hemisphere. Surgery was performed via the midline suboccipital approach. Similarly, we confirmed the vessels using ICG and the tumor was removed *en bloc*.

Conclusions: For hypervascular tumors of the posterior fossa, preoperative image assessment is important. Furthermore, the use of ICG during surgery is advantageous for surgical strategies where the feeding arteries and draining veins exist superficially in the operative field and are therefore easier to remove *en bloc*.



Key Words: Hemangioblastoma, hypervascular, indocyanine green, surgical videoangiography

INTRODUCTION

In high vascular tumors of the posterior fossa, surgical resection can be extremely challenging. Hemangioblastomas are common hypervascular tumors that occur in the posterior fossa. The prevalence of hemangioblastoma has been estimated to be approximately 1.5-2.5% of all intracranial tumors and 7-8% of posterior fossa tumors.^[1,8,9] Most lesions arise This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

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How to cite this article: Shinya T, Nagamine H, Sugawara Ki, Ishiuchi S. The usefulness of indocyanine green during surgery for hypervascular posterior fossa tumors. Surg Neurol Int 2018;9:90.

http://surgicalneurologyint.com/The-usefulness-of-indocyanine-green-during-surgery-for-hypervascular-posterior-fossa-tumors/

Surgical Neurology International 2018, 9:90

sporadically, although nearly one-third of patients have been diagnosed with von Hippel-Lindau (VHL) disease.^[3] Hemangioblastomas can be either predominantly cystic or solid tumors. These tumors are known to be highly vascularized with corresponding histological features of stromal cells strongly expressing vascular endothelial growth factor and abundant vascular cells.^[12] Treatment by surgical intervention may be necessary in certain cases, and removal of the tumors en bloc is important during surgery in order to minimize blood loss. However, because hemangioblastomas are frail and hypervascular, surgery carries a substantial risk for bleeding and resection can be difficult.^[19] Accordingly, tumor embolization before surgery has been considered to control bleeding during surgery, but there are a considerable number of reports detailing complications, such as brain swelling and/or intratumoral hemorrhage after embolism.^[4,6] Hence, the use of presurgical embolization has not been established as a standard practice.^[4,6] As another surgical strategy, several studies have reported on the use of indocyanine green (ICG) during surgery for tumor removal.^[7,8,10,17,19-21] However, ICG videoangiography is limited in that only tissue in the operative field is visible. When a deep-seated tumor is approached through a long, narrow corridor, it is not easy to clearly visualize tumor-related vessels with ICG videoangiography, and consequently, the technique may not be useful in the instance.^[10] For tumors of the posterior fossa, the surgical field is often deeper. Therefore, we report two cases in which the assessment of preoperative imaging with ICG could be advantageous from a surgical strategic viewpoint, where the feeding artery and draining vein exist superficially.

CASE HISTORY

Case 1

A 48-year-old male patient with a family history of VHL disease consulted an ophthalmologist because he had experienced blurred vision for 2 months prior to consultation. A diagnosis of bilateral retinal angioblastoma was made. Subsequently, magnetic resonance imaging (MRI) brain was performed to assess the patient for VHL. MRI showed a predominantly solid tumor with a maximum diameter of 3.0 cm accompanying an internal cyst close to the right cerebellopontine angle. In addition, edema extending to the brainstem was noted. The solid portion of the lesion was uniformly contrast-enhancing according to an MRI. Subsequent cerebral angiography identified feeding arteries from the superior cerebellar artery (SCA) and draining veins that converged onto the tentorial sinus [Figure 1]. The patient also exhibited deficits in working memory. We performed surgery because the tumor was compressing the posterior lobe of cerebellum and there was a decline in cognitive function.

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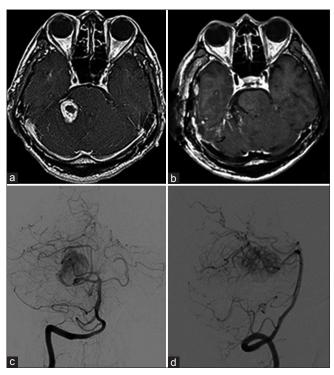


Figure I:The presurgical magnetic resonance (MR) image depicting an enhanced solid haemangioblastoma in the cerebellopontine angle (a). Postsurgical MR image depicting the resection of the tumour (b). Right vertebral artery angiogram exhibiting a hypervascular tumour near the superior cerebellar artery (c and d)

Surgery was performed via the posterior subtemporal transtentorial approach with the patient in the left supine lateral position. We intravenously injected mannitol and inserted a ventricular drainage tube into the supramarginal gyrus due to brain relaxation. After opening the dura mater, a surgical corridor was created by dissecting the bridging vein, and the tumor was confirmed by the retraction of the temporal lobe. The tumor was detached from the quadrigeminal bodies, followed by ICG fluorescence vascular angiography. First, the feeding arteries from the SCA were visualized, then the draining veins as they were converging with the tentorial sinus, and finally the transit arteries were carefully identified [Figure 2]. After careful assessment of the vasculature, the feeding arteries from the SCA were coagulated and cut. Finally, the draining veins were taken and the tumor was removed en bloc. The quantity of bleeding was approximately 640 ml. The pathological examination confirmed a hemangioblastoma World Health Organization (WHO) I.

Case 2

A 30-year-old woman patient visited our outpatient clinic after experiencing headache and blurred vision for 1 month. Her neurological examination was within the normal limits, except that bilateral congested papillae were identified. Her family history was unavailable because she was adopted. MRI revealed a solid tumor Surgical Neurology International 2018, 9:90

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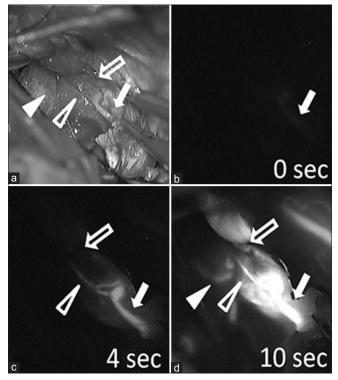


Figure 2: Surgical view (a) and indocyanine green (ICG) videography (b-d) during an operation. The main feeding arteries (arrow) are first filled with ICG (b), followed by transit feeders (open arrowhead), and then by simultaneous filling of drainers (open arrow) approximately 4 seconds after the main feeding arteries are visualized (c). Finally, non-feeding arteries (arrowhead) are filled with ICG approximately 10 seconds after the main feeding arteries (d)

with a maximum diameter of 3.5 cm accompanying an internal cyst in the left cerebellar hemisphere. In addition, extensive cerebral edema and ventricular dilation were noted. On presurgical brain MRI, the solid region showed uniform contrast enhancement. On cerebral angiography, feeding arteries were noted to arise from the left anterior inferior cerebellar artery (AICA) and left posterior inferior cerebellar artery (PICA), and both the draining veins to the petrosal vein and the inferior vermian vein were identified [Figure 3].

We performed 200 ml of 10% glycerol (twice a day) with dexamethasone (8 mg/day) for 6 days before the operation. Because the symptoms improved, we did not perform external ventricular drainage prior to the operation. Surgery was performed via the midline suboccipital approach with the patient in the prone position. The dura mater was dissected and ICG fluorescence angiography was performed. We observed the feeding artery coming off the PICA arose from the cerebellar hemisphere and drained via the horizontal fissure, additionally feeding vessels from the AICA were confirmed on the outside. The lesional veins drained into the inferior vermian vein [Figure 4]. Intraoperatively, the feeding artery from the PICA was coagulated first and then cut. Because there was a clear cleavage plane from the

surrounding tissue, which was not swollen, detachment was performed, leaving the thick draining veins intact. The most significant draining veins, which converged onto the petrosal vein, were positioned anteriorly on the outside. Finally, these veins were coagulated and cut, and the tumor was removed *en bloc*. The estimated blood loss was 500 ml. Pathological examination of the specimen confirmed hemangioblastoma WHO I.

DISCUSSION

Hemangioblastomas are benign tumors (WHO I) comprising neoplastic stromal cells and abundant small vessels.^[12] Surgical resection is the first line treatment, which is indicated in (a) symptomatic lesions, (b) urgency before symptoms develop, (c) lesions that are progressively enlarging on sequential scans, and (d) lesions that can or should not be treated with other modalities such as stereotactic radiosurgery.^[2,8,13] In the first case, we performed surgery because the tumor was compressing the posterior lobe of cerebellum, causing a decline in cognitive function.^[15,16] In the second case, surgery was performed because the patient exhibited signs of intracranial hypertension, such as headache and nausea. Hemangioblastomas have abundant blood flow comparable to arteriovenous malformations. The lesions are frail, and it can be difficult to control bleeding if removed in a piecemeal fashion, which also carries the risk of leaving some tumor behind,^[5] posing a high probability of recurrence. It has been widely advocated that these tumors must be removed *en bloc*.^[5,8,9]

In recent years, there have been several reports of presurgical embolization being performed in order to control bleeding during hemangioblastoma surgeries.^[3-6,9,11,14,18] However, serious complications such as stroke, hemorrhage, and swelling have also been reported,^[4,6] and therefore the procedure's utility has not been established.

Liu et al. compared the outcomes of groups in which presurgical embolization was or was not performed for cerebellar hemispheric hemangioblastomas. The authors reported that the duration of surgery was shortened, and the amount of blood loss and subsequent need for transfused blood were decreased in the embolization group.^[11] There are other studies reporting that fewer complications arose in cases where *n*-butyl-cyanoacrylate (NBCA) was used as the embolization material.^[9,14] However, this can result in recanalization after embolization because a tumor can develop collateral circulation from other vessels that embolized.[11] Particulate embolizing materials such as polyvinyl alcohol particles and embospheres are also widely used.^[4,6] However, there are a significant number of reports in which bleeding complications or swelling in the brain occurred after embolization with particular embolizing substances.^[4,6] There are other studies contesting the usefulness of presurgical

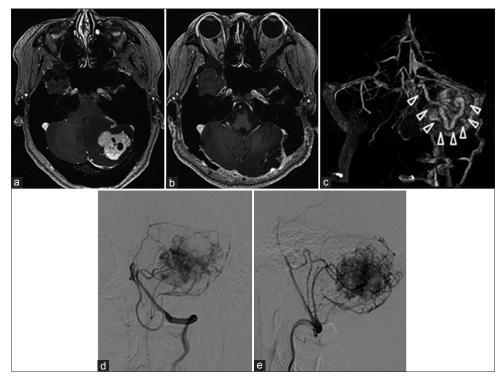


Figure 3: The presurgical magnetic resonance (MR) image demonstrating the enhanced solid haemangioblastoma in the cerebellum (a). The postsurgical MR image demonstrating the resection of the tumour (b). A Three-dimensional computed tomography angiogram during the late phase, exhibiting a draining vein (open arrowhead) flowing into the petrosal vein on the ventral side of the tumour (c). Left vertebral artery angiogram exhibiting a hypervascular tumour near the anterior inferior cerebellar artery and the posterior inferior cerebellar artery (d and e)

embolization. According to Ampie *et al.*, there are no significant differences in total tumor removal rate or the quantity of bleeding during surgery between embolized and nonembolized patients, but there is a high rate of complication associated with the embolism procedure itself.^[3] In addition, Takeuchi *et al.* reported swelling in the brain or cerebral hemorrhaging occurring in 70% of cases after partial embolization, rather than decreasing the risk of complications during surgery.^[18] Given that there are usually many feeding vessels in posterior fossa hemangioblastomas, complete embolization can be difficult and therefore, considerable care must be taken in order to avoid strokes.^[18]

Hemangioblastomas are characterized by abundant blood flow and it has been strongly advocated that they should be removed *en bloc*.^[5,8,9] Although evaluation of the respective vasculature is necessary before surgery in order to identify the anatomy of the relevant blood vessels, surgery can be made even safer if the blood vessels are visualized during surgery.

Hitherto, ICG has been employed routinely during surgery for brain aneurysms, cerebral arterial malformations, and bypass surgeries.^[7,17,19-21] Beyond that, the use of ICG for the identification of blood vessels in hemangioblastoma has previously been reported, with favorable results.^[7,8,10,17,19-21] According to Hojo *et al.*, ICG

can detect specific blood vessels via differences in the filling pattern.^[8] First, ICG fills the main feeding arteries and then transit arteries simultaneously with the draining veins. Finally, ICG fills the adjacent nonfeeding arteries.^[8]

However, the use of ICG also has its drawbacks. Deep blood vessels, covered by the cerebral parenchyma, cannot be visualized.^[7] In fact, there are usually many feeding vessels and they may be deep within posterior fossa hemangioblastomas.^[18] However, it can be considered that ICG could be useful in planning the surgical approach when using comprehensive assessment by presurgical angiography or three-dimensional visualization.^[22] In Case 1, we considered a suboccipital infratentorial approach, an occipital transtentorial approach, or a subtemporal transtentorial approach as alternative methods of approach. Both the suboccipital infratentorial approach and occipital transtentorial approach required excessive traction of the cerebellum and occipital lobe, and it was considered difficult to identify both the SCA and trochlear nerve in front of the tumor. However, the subtemporal transtentorial approach does not require excessive traction of temporal lobe, and it was possible to identify both the SCA and trochlear nerve in front of the tumor. In fact, we used ICG to confirm both the feeding artery and draining vein; therefore, the tumor was able to be removed en

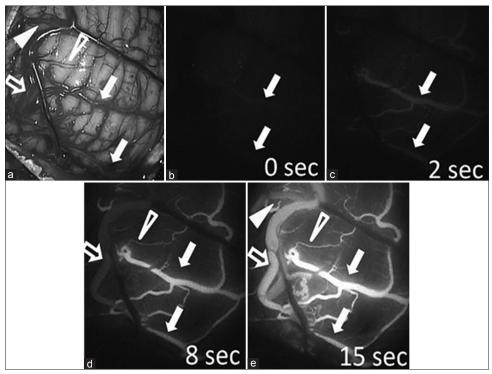


Figure 4: Surgical view (a) and indocyanine green (ICG) videography (b-e) during the operation. The main feeding arteries (arrows) are first filled with ICG (b and c), followed by transit feeders (open arrowhead) and the simultaneous filling of drainers (open arrow) approximately 8 seconds after the main feeding arteries are visualized (d). Finally, the non-feeding arteries (arrowhead) are filled with ICG approximately 15 seconds after the main feeding arteries are visualized (e)

bloc. In Case 2, because we used ICG to discover that the feeding arteries and the draining vein to the inferior vermian vein were on the surface, the surgery was able to be performed using the midline suboccipital approach. In this case as well, ICG was useful in making it possible to both control bleeding and remove the tumor *en bloc*, while preserving the normal blood vessels. However, it was impossible to identify the deep draining vein flowing into the petrosal vein during the initial administration of ICG, but it was assumed from the angiographic evaluation prior to surgery that the draining vein was in the outer basal region of the tumor.

We did not perform an embolization in either of these cases owing to concerns of the possibility of complications. Such complications could have been fatal (e.g., in Case #1) because of the proximity of the tumors to the brainstem. In such cases, we demonstrated that there is a way to use ICG without embolization. It seems that ICG can be successfully utilized by examining the surgical strategy with preoperative images.

CONCLUSION

For hypervascular tumors of the posterior fossa, preoperative image assessment is important. Furthermore, the use of ICG during surgery can be advantageous when the feeding arteries and draining veins exist superficially in the operative field and as a result, making tumor removal *en bloc* safer.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/ their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

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