



Cavernous Sinus capillary hemangioma: Case report and literature review

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ABSTRACT

Introduction: Capillary hemangioma of the cavernous sinus is considered a benign vascular skull base tumor. In this case report, a patient with capillary hemangioma located in the cavernous sinus who was operated on with endoscopic endonasal approach is presented.

Research question: What should be considered when planning the treatment for capillary hemangiomas located in the cavernous sinus?

Material and methods: The treatment approaches applied in 12 patients with a diagnosis of capillary hemangioma located in the cavernous sinus, reported in the English literature, are reviewed in this article. We presented the clinical and radiological results of the treatments.

Results: Cavernous sinus capillary hemangiomas are considered benign tumors. However, surgical treatment is preferred in the foreground because of the neurological deficits due to the compression effect caused by the mass. In the case reported by Masman et al. for the first time in the literature in 2021, they provided total excision with the endoscopic endonasal approach in the patient they first diagnosed by taking a biopsy with the endoscopic endonasal approach. In our case report, we provided total excision with endoscopic endonasal transsphenoidal approach.

Discussion and conclusion: Many factors should be considered when deciding on the treatment method in these patients. Although radiotherapy is quite effective in the treatment of capillary hemangioma, surgery should be preferred, especially in symptomatic patients. In patients who cannot undergo surgery, radiotherapy is very valuable, although the long-term results are not yet known.

1. Introduction

Capillary hemangiomas frequently involve the skin and soft tissues; with tumors often located on the face, scalp, chest, and back (Daenekindt et al., 2008; Simon et al., 2005). These can also involve the central nervous system, and when they do, the most common locations are the spinal cord and the cauda equina (Abe et al., 2004). Intracranial capillary hemangiomas, however, are rare. These can be found in different intracranial regions, one of which is the cavernous sinus. A capillary hemangioma of the cavernous sinus is considered a benign vascular skull base tumor (Grosu and Nieder, 2006). Radiologically, it can be confused with meningioma, hemangiopericytoma, metastasis and chondrosarcoma (Abe et al., 2004; Güneş et al., 2009; Karikari et al., 2006). Capillary hemangiomas located in the cavernous sinus have a risk of bleeding due to their

proximity to the internal carotid artery and a risk of neurological deficit due to cranial nerve compression.

There have been only 55 reported cases of intracranial capillary hemangioma in the literature. However, there are 10 publications reporting capillary hemangioma located in the cavernous sinus, and 12 cases are presented in these publications (Ishikawa et al., 2022; Low et al., 2019; Massman et al., 2021; Morace et al., 2012; Nawashiro et al., 2011; Pas et al., 2022; Phi et al., 2012; Saliba et al., 2009; Tsao et al., 2003). This case report presents a patient with a capillary hemangioma located in the cavernous sinus, who was operated on using an endoscopic endonasal approach.

Abbreviations: CT, Computed tomography; MRI, Magnetic resonance imaging.

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2. Case presentation

A 53-year-old male patient with no known underlying medical conditions presented to our clinic complaining of severe headaches that had started six months prior. The patient's headache occurred almost every day and often did not go away with medical treatment. A neurological examination of the patient revealed no neurological deficits. Due to a lesion observed in the right cavernous sinus on computed tomography (CT) (Fig. 1), contrast-enhanced brain magnetic resonance imaging (MRI) was ordered. The MRI revealed a mass-like lesion in the right cavernous sinus. The lesion appeared hypointense on T1-weighted images and hyperintense on T2-weighted images, showed no diffusion restriction, and enhanced homogeneously after the administration of gadolinium (Fig. 2). Vascular pathologies such as aneurysm, caroticocavernous fistula were excluded after MRI angiography and meningioma was considered in the preliminary diagnosis. (Fig. 3).

Following the completion of preoperative preparations, surgery was planned to use the endoscopic endonasal transsphenoidal approach. Preoperatively, a lumbar drain catheter was placed. During the surgery, bilateral nasal orifices were utilized to access the sphenoid sinus. The right internal carotid artery was identified, and the dura was opened medially. Advancing from the medial wall of the cavernous sinus, a gross total excision of the mass, which was adherent to the surrounding tissue, firm in consistency, and prone to bleeding, was performed (Fig. 4). The surgery was concluded with the placement of a synthetic dura and a nasal flap.

No bleeding was observed at the operation site in the postoperative CT (Fig. 5). Postoperatively, cerebrospinal fluid was drained through the lumbar drain catheter for three days, after which the catheter was removed. The patient, who only complained of headaches during the postoperative period, experienced complete resolution of symptoms following the removal of the lumbar drain catheter. After a 5-day postoperative observation period, the patient was discharged. No residual or recurrent mass was detected in the MRI performed at the 6th postoperative month of the patient, whose pathology result was reported as capillary hemangioma (Fig. 5). The follow-up of the patient, who did not receive any additional treatment after surgery, continues.

3. Discussion

In our case report, the symptoms, diagnosis, and treatment process of a 53-year-old male patient with a capillary hemangioma mass located in the cavernous sinus, who applied to us after complaining of headache, were explained. The ages of 12 patients with capillary hemangioma located in the cavernous sinus reported in the English literature were between the ages of 15–71, and 11 of the patients were female and only

one male. Although our patient was diagnosed with only headache, the patients reported in the literature had symptoms such as diplopia, vision loss, ophthalmoplegia, ptosis, facial pain, and facial hypoesthesia. These symptoms are due to the compression effect of the cavernous sinus mass and are not specific for capillary hemangioma. Although the CT and MRI findings of the cases reported in the literature are similar, it is possible to distinguish them from other intracranial masses only histopathologically. Also, this article reviews the treatment approaches applied to patients diagnosed with capillary hemangioma located in the cavernous sinus (Table 1).

Tsao et al. (2003) reported on two patients with capillary hemangioma located in the cavernous sinus. In both cases, SFRT was chosen as the treatment method following a biopsy-confirmed diagnosis. Post-radiotherapy MRI scans revealed no contrast-enhanced tumor tissue in one patient, while minor residual tumor tissue was observed in the other patient despite a reduction in tumor size (Tsao et al., 2003).

In 2006, Grosu et al. reported on a patient who had previously undergone multiple surgeries and experienced tumor recurrence. After SFRT, the tumor volume decreased and clinical symptoms regressed (Grosu and Nieder, 2006).

In 2009, Saliba et al. carried out the first macroscopic total excision of the transcranial tumor using a subtemporal approach. It has been reported that the symptoms of the patient regressed after the surgery (Saliba et al., 2009).

Nawashiro et al. (2011) reported on a patient presenting with diplopia and periorbital pain who was diagnosed by biopsy using an endoscopic transsphenoidal approach. However, their article did not specify whether the patient received additional treatment or provided follow-up information. It is inconsistent with the clinical follow-up and treatment conditions reported in our and other articles in the literature.

Phi et al. (2012) reported on four patients with intracranial capillary hemangioma, including a patient with a cavernous capillary hemangioma. Postoperative SFRT was administered to a patient who underwent partial tumor excision using a microscopic transsphenoidal approach, and a decrease in tumor size was observed during follow-up (Phi et al., 2012).

Morace et al. (2012) shared their experiences with two patients who had capillary hemangiomas extending from the cavernous sinus to the anterior temporal region in their article, which described four patients with intracranial capillary hemangiomas. In both cases, subtotal excision was performed using a frontotemporal approach, followed by SFRT. Reduction in tumor size was observed post-radiotherapy (Morace et al., 2012).

Low et al. (2019) reported on a patient who initially underwent a biopsy with the endoscopic endonasal transsphenoidal approach, and when the diagnosis could not be confirmed, a second biopsy and

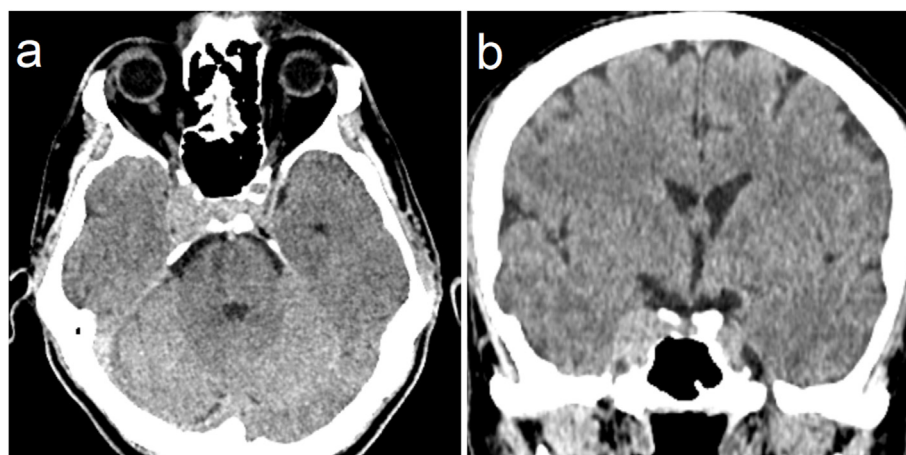


Fig. 1. CT imaging of the right cavernous sinus capillary hemangioma in axial (A) and coronal (B) sections.

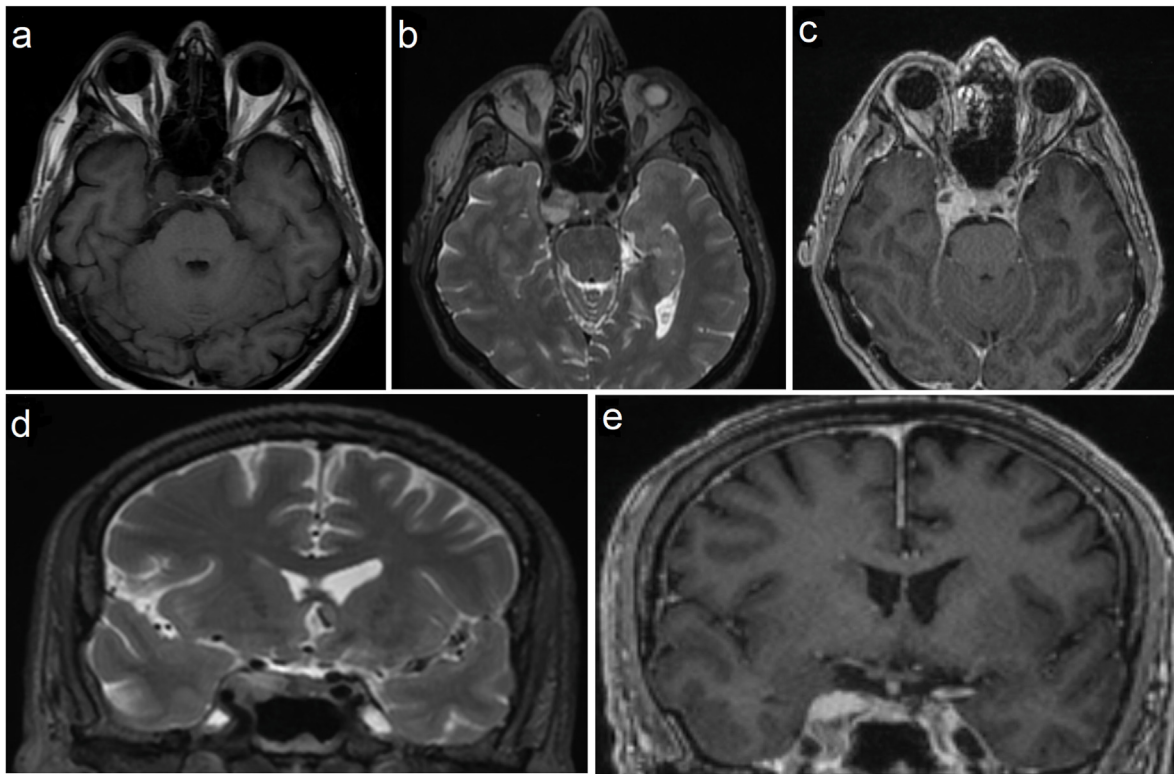


Fig. 2. On magnetic resonance imaging, capillary hemangioma is hypointense on T1 sequence axial section (a) and hyperintense on T2 sequence axial (b) and coronal section (d). Capillary hemangioma appearing homogeneously enhanced on axial (c) and coronal (e) sections on Gadolinium enhanced T1 sequence images.

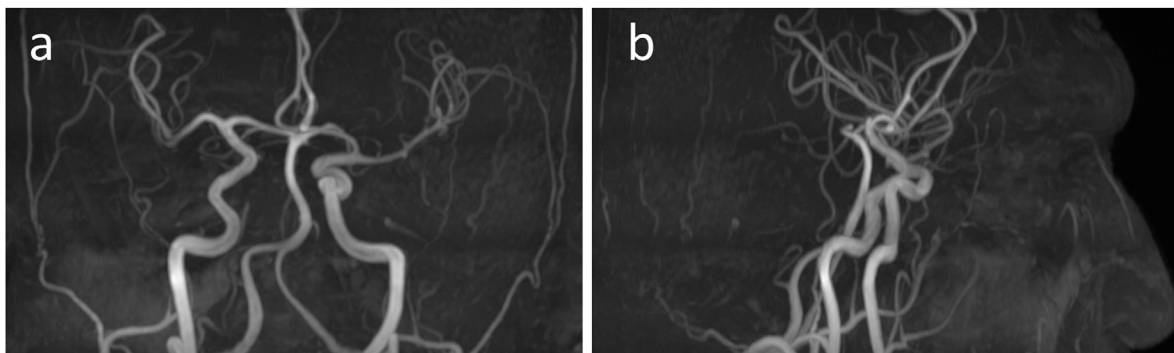


Fig. 3. MRI angiography imaging of the patient a) anterior posterior b) lateral.

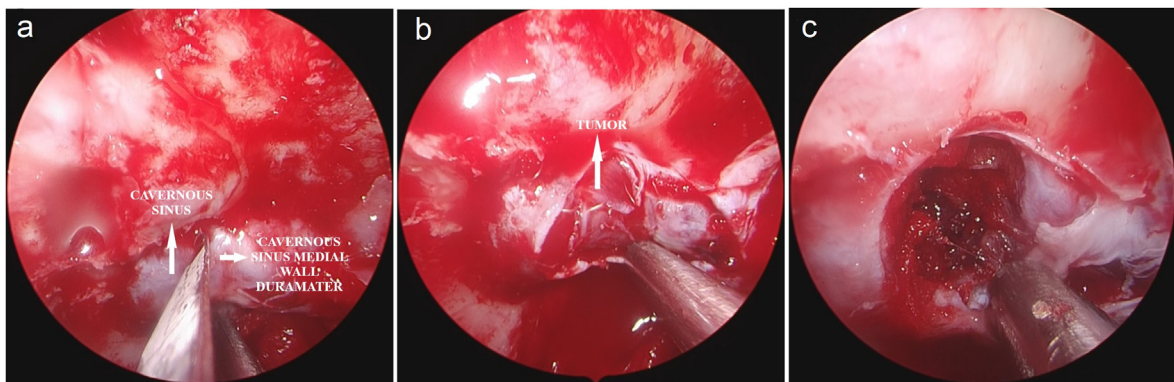


Fig. 4. In endoscopic endonasal transsphenoidal approach a: Cavernous sinus dura incision after removal of the bone above the sella b: Appearance of cavernous sinus localized mass after dura opening c: Operation site after gross total resection.

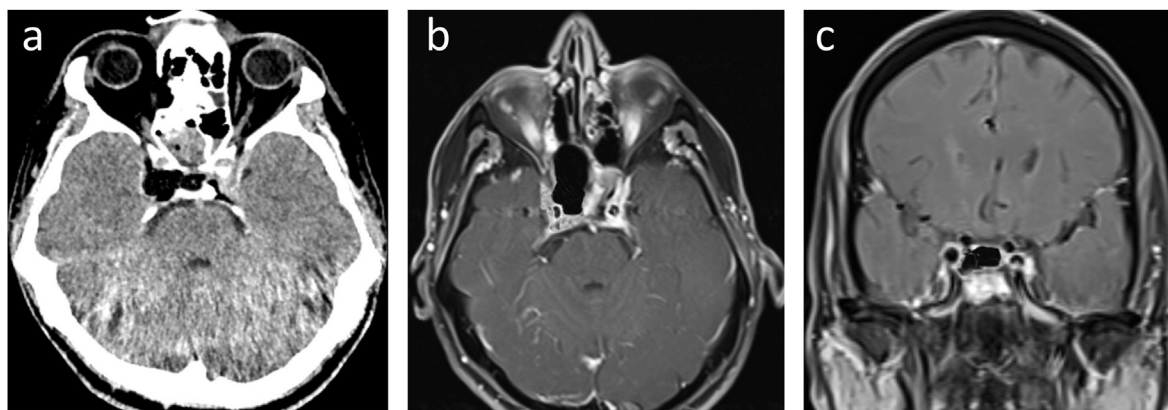


Fig. 5. Postoperative imaging after tumor excision with endoscopic transsphenoidal approach a) Axial section b) axial image on contrast enhanced T1 MRI c) Coronal image on contrast enhanced T1 MRI.

Table 1

Summary table of capillary hemangioma cases located in the cavernous sinus reported in the literature CS:Cavernous sinus, EEA:Endoscopic endonasal approach, F:Female, L:Left, M:Male, NS:Not specified, R:Right, SFRT:Stereotactic fractionated radiotherapy.

Author and Year	Age (years)	Sex	Location	Symptom	Surgical Approach	Resection Degree	Radiotherapy/ Medication	Outcome
Tsao et al. (2003)	15	F	R CS	Diplopia and proptosis	Frontotemporal	Biopsy and partial resection	SFRT	Local tumor control (18 months)
	19	F	L CS	Diplopia	NS	Biopsy	SFRT	No recurrence (21 months)
Grosu and Nieder (2006)	62	M	R CS, extending to orbita and suprasellar region	Epilepsy, complete visual loss of the right eye, paresis of the right oculomotor nerve and both decreased visual acuity and fields of the left eye	Frontotemporal craniotomy	Subtotal	Postop SFRT	Local tumor reduction 3 prior surgeries with recurrence
Saliba et al. (2009)	60	F	R CS extending to foramen ovale & petrous ICA	Chronic facial pain and paresthesia	Subtemporal craniotomy	Total		Symptom resolution
Nawashiro et al. (2011)	65	F	L CS	Diplopia, left total ophthalmoplegia	EEA	Biopsy		Spontaneous involution
Phi et al. (2012)	44	F	R CS	Visual field loss	Transsphenoidal	Partial	Postop SFRT	Tumor reduction (23 months)
Morace et al. (2012)	26	F	R CS	Galactorrhea and irregular menstrual cycles	R frontotemporal craniotomy	Subtotal	Postop SFRT	Local tumor control (3 years)
	61	F	L CS	Left eye visual impairment and a left second trigeminal branch sensory deficit.	L frontotemporal craniotomy	Subtotal	Postop SFRT	Local tumor control (11 month)
Low et al. (2019)	64	F	L CS	Headache, ptosis, paresthesia	EEA, pterional craniotomy	Biopsy, total	Dexamethasone	NS
Massman et al. (2021)	23	F	R CS	Ophthalmoplegia, ptosis, diplopia, facial pain, hypoesthesia	EEA, EEA	Biopsy, total		No recurrence (1 year)
Ishikawa et al. (2022)	28	F	R CS	Diplopia	EEA	Total		No recurrence (1.5 year)
Pas et al. (2022)	71	F	L CS	Headache, diplopia	EEA	Partial		Local tumor control (3 months)
Present Study	53	M	R CS	Headache	EEA	Total		No recurrence (6 months)

debulking were performed using a pterional approach. However, the article did not include any follow-up information for this patient (Low et al., 2019).

In a case reported by Massman et al. (2021), the first of its kind in literature, a biopsy was initially performed using an endoscopic endonasal approach. Following the biopsy and confirmation of diagnosis, total excision was achieved using the same endoscopic endonasal approach. No recurrence was observed in the MRI performed after 1 year of patient follow-up. (Massman et al., 2021).

Pas et al. (2022) reported a case in which an endoscopic endonasal transpterygoid approach was used for surgery, and Ishikawa et al. (2022) reported a case where total excision was achieved using an endoscopic

endonasal transsphenoidal approach (Ishikawa et al., 2022; Pas et al., 2022). In our case report, we also achieved total excision using the endoscopic endonasal transsphenoidal approach. Despite the patient not showing any deficits in the neurological examination, we opted for surgical intervention due to his severe headache.

As revealed in our literature review, we chose the endoscopic endonasal transsphenoidal approach to the cavernous sinus for surgical planning, an approach that has gained increased use recently. Compared to other transcranial and microscopic transsphenoidal approaches, the endoscopic endonasal approach is safer as it provides a broader view of the medial wall of the cavernous sinus (Ceylan et al., 2010; Fernandez-Miranda et al., 2018; Woodworth et al., 2014).

4. Conclusion

When deciding on the treatment method for these patients, several factors should be considered. While radiotherapy is highly effective in treating capillary hemangioma, surgery is generally the preferred option, particularly for symptomatic patients. For those unable to undergo surgery, radiotherapy remains a valuable treatment option, even though its long-term results are yet to be fully understood.

Both transcranial and endoscopic approaches may be considered when operating on a capillary hemangioma located in the cavernous sinus. However, in centers with a skilled skull base team, endoscopic approaches are typically superior to transcranial ones. It's important to note that if the tumor adheres to crucial neurological and vascular structures during surgery, radiotherapy may be employed as a complementary treatment.

Informed consent

Written informed consent for submission of the paper was obtained from the patient.

Disclosure statement

The author declares no conflict of interest.

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There are no additional relationships to disclose.

There are no additional activities to disclose.

Ethical approval

Patient was consented for approval for writing the case and publication.

Contributors

All authors have contributed to the article and have approved the final article.

Structured review

The treatment approaches applied in 12 patients with a diagnosis of capillary hemangioma located in the cavernous sinus, reported in the English literature, are reviewed in this article. Tsao et al. (2003) reported 2 patients with capillary hemangioma located in the cavernous sinus. Radiotherapy was preferred as the treatment method in both patients after the diagnosis was made by biopsy. In the MRI of the patients after radiotherapy, no contrast-enhanced tumor tissue was found in one patient, while minor residual tumor tissue remained in the other patient despite the reduction in tumor size (Tsao et al., 2003). In 2006, Grosu et al. reported that after fractionated stereotactic radiosurgery performed in a patient who had previously undergone multiple surgery and had recurrence in the follow-up, tumor volume was reduced, and clinical symptoms regressed (Grosu and Nieder, 2006). Saliba et al. on the other hand, performed the first macroscopic total excision of the transcranial with the subtemporal approach in 2009 (Saliba et al., 2009). Nawashiro et al. (2011) reported on a patient who presented with diplopia and periorbital pain, diagnosed by biopsy with the endoscopic transsphenoidal approach, but in their article, whether the patient was given additional treatment or follow-up information was not specified.⁹ Phi et al. (2012) reported 4 patients with intracranial capillary hemangioma and included a patient with cavernous capillary hemangioma. Radiotherapy was applied in the postoperative period in the patient who underwent partial tumor excision with microscopic transsphenoidal approach. A decrease in tumor size was observed during follow-up (Phi et al., 2012). Morace et al. (2012) also shared their experiences with 2 patients with capillary hemangioma extending from the cavernous sinus

to the anterior temporal in their article describing 4 patients with intracranial capillary hemangiomas. In both patients, radiotherapy was applied by providing subtotal excision with frontotemporal approach. A reduction in tumor size was observed after radiotherapy (Morace et al., 2012). Low et al. (2019) reported a patient who underwent biopsy and debulking with the pterional approach, after first taking a biopsy with the endoscopic endonasal transsphenoidal approach, after which the diagnosis could not be made. The patient's follow-up information was not included in the article (Low et al., 2019). In the case reported by Masman et al. for the first time in the literature in 2021, they provided total excision with the endoscopic endonasal approach in the patient they first diagnosed by taking a biopsy with the endoscopic endonasal approach (Massman et al., 2021). Pas et al. reported a patient who operated with endoscopic endonasal transpterygoid approach (2022) and Ishikawa et al. (2022) operated with endoscopic endonasal transsphenoidal approach and performed total excision (Ishikawa et al., 2022; Pas et al., 2022). In our case report, we provided total excision with endoscopic endonasal transsphenoidal approach. Although there was no deficit in the neurological examination, we considered surgical intervention in the patient who had severe headache. As noticed in our literature review in surgical planning, we preferred the endoscopic endonasal transsphenoidal approach to the cavernous sinus, which has been increasingly used recently. The endoscopic endonasal approach is safer than other transcranial and microscopic transsphenoidal approaches because it provides a wider view of the medial wall of the cavernous sinus (Ceylan et al., 2010; Fernandez-Miranda et al., 2018; Woodworth et al., 2014).

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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