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Endoscopic Endonasal Approach to Cavernous Sinus Hemangioma: A Report of Two Cases and Review of the Literature

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ABSTRACT

Cavernous sinus hemangiomas (CSHs) are rare, benign, slow-growing neoplasms within the cavernous sinus. Laterally located to these lesions, the cranial nerves and carotid artery are subject to injury during removal of hemangiomas through a transcranial route. Therefore, for surgical management of cavernous sinus hemangiomas a medial corridor granted through an endoscop- ic endonasal approach may be less traumatic to the neurovascular bundle.

Case-1 describes a 23-year old male who presented with intermittent blurred vision and very mild ptosis on the right side for two years before admission. Fundoscopic exam, visual acuity and perimetry tests were normal. With intense enhancement after contrast administration, both brain MRI and CT scan demonstrated an extra-axial mass in the right cavernous sinus.

Case-2 presents a 59-year-old male, a known case of renal oncocytoma who underwent nephrectomy a year before, with chief complain of moderate intermittent headaches. Imaging study of the brain by MRI revealed a sellar mass.

Near-total resection for case-1 and gross total resection for case-2 were performed through the an extended endoscopic endonasal approach.

We report two cases of near-total and gross total resection of CSHs via an extend- ed endoscopic endonasal approach substantiating advances in minimal access neurosurgery to the treatment of such grim lesions of an intricate vicinity of the skull base. At the same time, we focus to review extensively the growing yet heterogenous literature of the past twenty years on the broached topic. The evolution of extended endoscopic endonasal approach over the past two decades changes the dynamism of the surgical practices steeped in tradition and provides a safer alternate route for preserving cranial nerves of this anatomic region.

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Introduction

Cavernous sinus hemangiomas (CSHs) are uncommon, benign tumors, accounting for approximately 2% of all pathologies within the dural confines of the cavernous sinus. Indolent nature of these lesions leads to acute or subacute ophthalmoparesis, followed by facial hypoesthesia, visual symptoms and hypopituitarism.

Proximity to the neurovascular structures and predisposition to massive hemorrhage during surgery intensify the perils of interventions for CSHs. Mainly, they occur as a single lesion in middle-aged individuals and more frequently found in females which may predict the importance of estrogen in their development [21]. In current approaches to CSHs, microsurgical resection, interventional embolization, fractionated radiotherapy and radiosurgery are routinely engaged [2, 19, 22].

Although various approaches including craniotomy, microscopic transsphenoidal partial resection and endoscopic endonasal tumor resection have been employed, micorsurgery of CSHs has remained open to the challenge of any curious research. Emplacement of cranial nerves lateral to the cavernous sinus and carotid artery and associated risk of their injury during surgery authoritatively imposes a more judicious medial approach, found to be less traumatic.

Inevitable progress of endonasal approaches may be traced out in the development of instrumentation, techniques and resection nuances for endocscopic skull base surgery. While cavernous sinus hemangiomas have been addressed by endoscopic surgeons for more than a decade now, there has been a full-scale resurgence of attention in advancing this field by more systematic methods. Central to these efforts is comparison of techniques and outcomes across institutions to establish consensus and extend application of endonasal endoscopic approach to perceivably-suitable CSHs. Cognizant of these remarks, we present two patients with CSHs who underwent endoscopic endonasal tumor resection, elaborated with a brief review of the pertinent literature.

Case presentation

Case 1

A 23-year-old male with no significant past medical history presented with transient blurred vision and mild ptosis for two years before admission. Fundoscopic examination, visual acuity and perimetry were found to be normal. CT scan and MRI revealed an extra-axial mass with intense enhancement after contrast injection in size 29*24*18 mm in the right cavernous sinus (Fig.1A-D). Pituitary hormones values were within normal range. For definite diagnosis we decided to take a biopsy by endoscopic endonasal transsphenoidal approach. Following the removal of the anterior wall of the sella, the dura was incised to expose the lesion. At this stage, a highly vascular mass was encountered with a gross appearance of hemangoima or angiomatous meningioma, a biopsy was taken.

Histological examination revealed a cavernous sinus hemangioma (Fig. 1E). A week later, an expanded endoscopic endonasal approach was attempted, the anteriour surface of the right cavernous sinus was exposed. Due to adherence of the lesion to lateral wall of the CS and internal carotid artery, sub-total removal of the tumor was done (Fig.1F). A mild CSF leakage occurred. The dura was solely repaired by a fascia lata graft. Moreover, a fat graft was interposed between the residual tumor and normal pituitary gland in case of any future need for adjuvant radiation therapy.

Following the surgery, the patient reported improvement in spells of blurred vision. Neurological examinations and hormonal profiles did not reveal any defects or abnormalities.. Postoperative MRI with gadolinium enhancement indicated the sharp margin of residual tumor on the lateral wall of the right cavernous sinus adhered to the ICA (Fig.1G&H). After 42 months of follow-up, the patient remained asymptomatic and the imaging findings showed no tumor growth.

Case 2

A 59-year-old male with a history of renal oncocytoma, for which he underwent nephrectomy a year before, presented with headache and brain MRI finding of a sellar mass of 27*21*20 mm in size (Fig. 2A-D). Pituitary hormonal profile was normal at the time of presentation. Although metastasis workup indicated no abnormality, a metastasis of the original tumor came first in our differential diagnosis. Therefore, an extended endonasal transsphenoidal approach was utilized to expose the sella and the right cavernous sinus.

Subsequent to incision of the dura, a brownish-redand non-suctionable mass was detected. While metastatic renal oncocytoma was initially entertained, cavernous sinus hemanioma was speculated as a

more likely diagnosis while gross total resection of the tumor was successfully achieved (Fig. 2E). Over the course of the operation, 1800 ml blood loss was incurred largely due to the cavernous sinus walls and not the tumor itself. The dura matter was repaired by an onlay fascia lata graft. Histological examination of the specimen verified presumptuous diagnosis of CSH (Fig. 2F). Tracking the patient after the surgery for 36 months, no abnormality in pituitary hormone levels was found. Post-operative MRI at 3, 12, 36 months did not bring in to display any residual or recurrent tumor (Fig. 2 G&H). Of note, the patient renal oncocytoma is still in remission without any evidence of recurrence.

Discussion

Cavernous sinus hemangiomas (CSHs) are the uncommon, highly-benign, vascular lesions of skull base responsible for approximately 2% of all pathologies within the dural boundaries of the cavernous sinus [9].

Rare cases of intrasellar cavernous hemangiomas have been reported which may be entertained in differential diagnosis of pituitary adenomas and meningiomas [4, 5, 10, 11]. Though they are benign, these slow-growing lesions can thrive within the cavernouns sinus invoking a mass effect, producing clinical symptoms and signs which may consist of headache, ocular pain, cranial nerve deficits, ophthalmoparesis and less commonly, endocrinopathies, exophthalmus or trigminal neuralgia [11, 25]. While expansion of the tumor medially encloses the internal carotid artery (ICA) or presses the pituitary gland, tumor may burgeon superiorly toward the optic chiasm, dislocating the subarachnoid ICA or laterally compress the temporal lobe [15].

Our case number 1 presented with mild ptosis but no other cranial nerve abnormality or endocrinopathy. His prolactin level at the time of presentation was 10.2 ng/ml. Whereas case number 2 was totally asymptomatic and the metastatic workup for the primary tumor revealed the cavernous sinus involvement and normal prolactin level. Table1 which reviews the literature over the past 20 years indicates headache, visual field deficits and diplopia as the most common clinical presentation of hemagioma of the cavernous sinus, respectively.

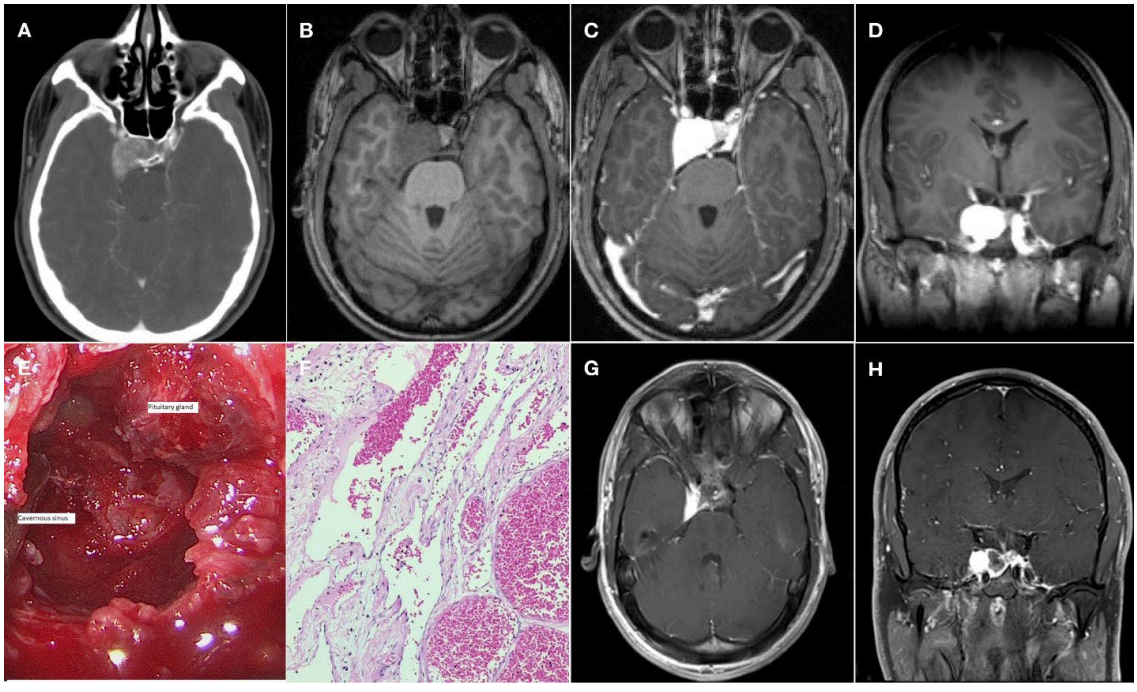

Fig. 1. A-B: Preoperative Post contrast axial CT scan, and coronal gadolinium-enhanced magnetic resonance imagings without and with gadolinium injection demonstrate a mass with intense enhancement within the lateral sella and cavernous sinus.
C: Postoperative coronal gadolinium enhanced MRI scans demonstrate near total resection with a residual tumor adhered to the internal carotid artery ad lateral wall of cavernous sinus.
D: Intraoperative endosopic image of endonasal approach to the right cavernous sinus.
E: Photomicrograph of the histopathologic examination demonstrating multiple dilated vascular channels that are lined with endothelium and separated by a matrix of fibrous tissue. H & E, original magnification ×100.

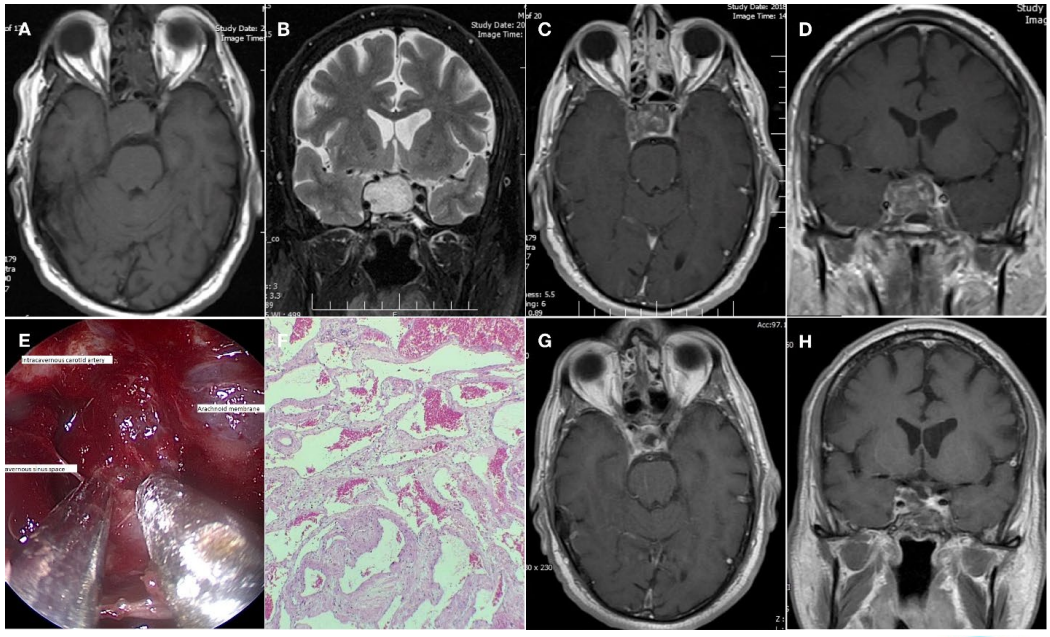

Fig.2. Contrast-enhanced T1-weighted coronal MR images(A) demonstrating a mass on the right side of sella with non-homogeneous enhancement.
B: Postoperative coronal T1-weighted MR imaging reveal gross total tumor resection with preservation of normal pituitary gland.
C: Intraoperative image showing a close up view of endoscopic endonasal transsphenoidal approach to the right cavernous sinus after gross total tumor resection.
D: Histopathologic examination with H&E stain shows multiple large vascular channeles lined by bland-looking endothelial cells which are filled with RBCs. Original magnification ×40.

These lesions typically appear as hypo or isointense on T1-weigted images and hyperintense on T2-weighted MR images; which alone lend a differential diagnosis of meningioma, schwannoma, pituitary adenoma, or CSH [3, 23]. These findings differ from those of meningiomas, which usually exhibit a low-intensity signal on T2-weighted images, however, they match MRI characterization of neurinomas [8]. Linskey et al. reported that 80% of cavernous hemangiomas in the cavernous sinus display a blush or blood pool, described as “flecked” in appearance [11]. The lesions are chiefly fed by the meningohypophyseal trunk and cavernous branches from the internal carotid and middle meningeal arteries [8]. Preoperative imaging for case number 1 could not explicitly rule out a diagnosis other than hemangioma, for example a meningioma. So, we had to take an endoscopic biopsy to confirm the diagnosis through histopathology. Unfortunately, the frozen section was not diagnostic. As to our case number 2, our primary impression was a metastatic lesion, other pathologies including a pituitary adenoma or a hemangioma could not be excluded though.

To differentiate CSHs from other pathologies, Tc-99m RBC scintigraphy is primarily suggested which conspicuously provide this tracer within the cavernous sinus, a finding specific to CSHs and not evident in other extra-axial masses in this region [3]. Preoperative tumor assessment requires precise diagnostic radiologic studies to fully determine dimensions and relationship of tumor to the neighboring neurovascular structures [1].

Treatment strategies currently practicable for CSHs can be divided into two main surgical and nonsurgical categories. Surgical interventions entail transcranial and endoscopic endonasal approaches, while Gamma knife and fractionated radiotherapy are considered nonsurgical methods [3]. Besides, treatment schemes are individualized commensurate with size, invasion and expansion, hemorrhagicity of tumor, patient’s desire, as well as the surgeon’s skills and available facilities. However, according to published articles, surgical treatments are essentially recommended for giant masses while small to medium size lesions may be best dislodged by non-surgical tactics like gamma knife radiosurgery [24]. In comparison, surgical approaches are viewed as more curative by virtue of capability for total resection in association with less relapse rate.

Table 1. Studies which used endoscopic endonasal surgery for cavernous sinus hemangioma

Source	Age (Yr)/ Sex	Initial Tumor Size (mm) or Volume (cm^3)	Clinical Presentation	Extent of Resection	Postoperative Complications	Latest Follow-Up
Fraser 2008 [9]	50/M	17×16×28 mm	Dizziness Severe Headache Nausea & Vomiting Mild Diplopia	GTR	None	?
Ma 2014 [16]	?	35×18×27 mm	Blurred Vision ↓ Libido Bitemporal Headache	STR	?	?
Patrona 2017 [20]	?	5.16 cm^3	Headache	GTR	None	None
Patrona 2017 [20]	?	25.77 cm^3	CN V2 Palsy	No Change	None	No Change
Patrona 2017 [20]	?	8.69 cm^3	CN VI Palsy	GTR	None	Resolved CN Palsy
Nishimura 2018 [17]	49/F	5.12 cm^3	Headache CN VI Palsy Diplopia	GTR	None	No Change
Khattar 2018 [13]	69/M	27×19×17 mm	Facial Pain Nocturia ↓ Libid0	GTR	None	None
Noblett 2018 [18]	57/M	35×41×24 mm	Headache	STR	None	Radiographic progression Mild Ptosis, Mild Facial Dysesthesia after 18 months
Das 2018 [7]	66/F	?	Headache Bitemporal Hemianopsia	PR	?	?
Li 2018 [14]	40/F	24 mm	Headache ↓ Vision No Menstruation Mild Diplopia	PR	?	?
Li 2018 [14]	36/F	11	↓ Vision	GTR	?	?
Li 2018 [14]	66/F	35 mm	↓ Vision Facial Hypesthesia	PR	?	?
Li 2018 [14]	54/F	13mm	Diplopia	GTR	?	?
Current Study	23/M	29×24×18 mm	Mild Ptosis Intermittent Visual obscuration	STR	None	Stable Tumor residue
Current Study	59/M	27×21×20	Asymptomatic	GTR	None	None

?: Data not published; GTR: Gross Total Resection; STR: Sub-Total Resection; PR, Partial Resection

Despite relentless advances in microsurgery techniques over the past two decades, evident by declining mortality rates, the mere event of hemorrhagicity complicates the key thread of the method and poses a stern challenge [18] [24]. Controversy still exists over the preferred management and selection of a gold standard lingered in equivocality. However, the surgical approaches generallyhave remained the first-choice treatment [8].

Among surgical options, different approaches including the frontotemporal, transsylvian, lateral wall, extradural subtermporal and microscopic transsphenoidal have been employed, many of them, with crippling limitations that temper enthusiasm of their use. However, march of new advances in mirosurgical techniques led Linskey and Sekhar to accomplish successful total resection in three patients with CSHs [15].

In addition to its application in the resection of pituitary adenomas outstretched intracavernously, endonasal endoscopic approach is favored to alternative interventions for CSHs since the tumor extension is prominently lateral to the sella with significant involvement of the middle fossa [6, 9]. The current report, to our knowledge, is the largest systematic study on the results of endoscopic endonasal surgery on hemangiomas within the cavernous sinus with or without sellar involvement which can be seen in table 1.

The endoscopic approach permits to identify, less injuriously, the normal pituitary gland, cavernous sinus and intracavernous ICA [9]. Furthermore, dynamic visualization and accessibility from medial to the lateral wall of cavernous sinus decreases the risk of cranial nerve injury and safeguard against brain retraction associated with transcranial approaches [12].

Rising interest in endoscopic endonasal transsphenoidal approach ensued once its promising outcomes were more frequented with the first total resection by Fraser et al. The other cases of CSHs or intrasellar hemanigoma who underwent endsocpic endonasal surgery were Cobbs and Jeon et al. pressed to confide entirely in subtotal resection since Noblett and Wilson studies advised against GTR which may cause a perilous dural tear and was liable to a marked hemorrhage [5, 11, 18]. However, Fraser group with reliance on endoscopic advancements, more sophisticated scope tools and resection nuances profess GTR as safe and recommendable [9]. Patrona group, who studied a larger case series of nonadenomatous and nonmeningeal tumors of cavernous sinus including hemangioma, achieved GTR in two of three CSHs and reported they were not able to retain homeostasis in the third case [20]. Moreover, Nishimura et al. presented a case report of CSHs in 2018 for which they performed a GTR with consequent recurrence of the tumor and advocate exercising radiosurgery as adjuvant or primary treatment [17]. A case presentation of CSH done by Indiana university in 2018 reaffirmed endoscopic endonasal surgery as the most favorable procedure in the current armamentarium to treat this nuisance while they reported a CSF leak and no serious bleeding [13]. Noblett group at Davis in 2018 while remained equivocal on choice of optimal approach, on a cautionary note, favored subtotal resection for most patients with CSHfollowed by radiosurgery for long-term control [18].

In our case-1, anatomy of the tumor medial to the cavernous sinus with no significant extension in the sella rendered our patient a suitable candidate for endonasal endoscopic resection which allowed us to explore the cavernous sinus and its proximities, the sella and intracavernous ICA, as well as a medial access to the cavernous sinus with no deterrent cranial nerve before resection of the tumor. However, the tumor's adherence to the lateral wall of the cavernous sinus and ICA discouraged us from doing GTR and we had to confide in subtotal resection. However, in our case-2, we successfully achieved GTR with no long-term complication such as CN deficit or tumor recurrence. Table 1 (2008-2020) substantiates the practicability of GTR in CSHs through endoscopic approach, eleven cases of GTR (8) and STR (3) out of fifteen (73%) with no significant negative outcome.

In the two cases reported in this study, proximity of the tumor to the sella and their medial extension from the lateral wall of the cavernous sinus to the midline justified an endonasal approach far more prudent for allowing to identify the intact pituitary gland, the cavernous sinus and the intracavernous ICA. Embracing endonasal approach to the cavernous sinus, evident in our two cases and other studies according to table 1, represent a paradigm shift in surgical practices, assumed to treat CSHs, by granting more pragmatic accessibility, not restrained by bullying vulnerable lateral-wall elements and enhancing prognostic implications.

Conclusion

Cavernous sinus hemangiomas are rare benign neoplasms which may be misdiagnosed as pituitary adenomas, meningiomas, metastases or

schwannomas. This tumor traditionally treated with an open craniotomy. With the evolution of extended endoscopic endonasal approach since two decades ago, this corridor could be a safe and judicious alternative which was successful and this approach meaningfully enhance the current armamentarium for surgical practices of CSHs. The significant outcomes of our challenging cases favor endoscopic endonasal transsphenoidal procedure and provide us with more tenable grounds to more broadly adopt and more intensely probe this laboriously progressive portal

Ethical Considerations

This research has been performed in accordance with WMA declaration of Helsinki

Consent

No information which leads to identification of the presented patients was included in this paper

Compliance with ethical guidelines

There were no ethical considerations to be considered in this research.

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Authors' contributions

All authors equally contributed in preparing this article.

Conflict of interest

No conflict of interest or competing interest exists in the submission of this manuscript and all authors approved of its publication

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