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Case Report

Multiport combined endoscopic endonasal and transorbital approach to orbital schwannoma

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ABSTRACT

Background: We present a case report describing the use of combined endoscopic endonasal and transorbital approach (EETOA) for intraorbital schwannoma that grew rapidly during pregnancy.

Case Description: A 27-year-old woman who presented with headache was incidentally diagnosed with a tumor mass 1 cm in diameter near the right superior orbital fissure. Treatment of the tumor was deferred to follow-up. One month later, the patient became pregnant, and in the last trimester, she developed right abducens palsy and mildly dilated pupil. Magnetic resonance imaging (MRI) showed that the tumor mass had grown rapidly with a maximal diameter of 5 cm and had extended into the orbit through the superior orbital fissure. After spontaneous vaginal delivery, EETOA was planned to remove both the intraorbital region and cavernous sinus compartment to avoid craniotomy. Surgical resection began with an endoscopic endonasal approach. Orbital decompression was performed by removing the lamina papyracea, and the tumor was resected in the lateral cavernous sinus compartment. An eyebrow incision was made, and endoscopic transorbital surgery was performed to remove the intraorbital region. Multi-perspective views during EETOA enabled gross total resection of the tumor and confirmed by intraoperative MRI. The pathological diagnosis was schwannoma. The patient's abducens nerve palsy improved after surgery.

Conclusion: EETOA can offer maximal exposure and resection for tumors extending from the cavernous sinus to the orbit without craniotomy in selected cases.

Keywords: Combined endoscopic endonasal and transorbital approach, Multiport surgery, Pregnancy, Schwannoma

INTRODUCTION

The orbit is an anatomically complex area, and intraorbital tumors can be approached from different surgical corridors.[32] Since the eyeball plays a central role in facial expressions, treatment strategies must consider not only the effective resection of the tumor but also cosmetic factors.

The evolution of the endoscopic endonasal approach (EEA) has enabled the removal of tumors that would have required craniotomy in the past.[7,15] Several multiport skull base approaches have recently been proposed.[9,22] For instance, the addition of the transorbital route to the endonasal approach has been reported to improve access to the skull base tumor in a cadaveric study[2,4,5,11,13] and in clinical settings.[5,13,14,37]

In this report, we present a case of intraorbital schwannoma that grew rapidly during pregnancy. After spontaneous vaginal delivery, the multiport combined endoscopic endonasal and transorbital approach (EETOA) was successfully performed to remove the tumor completely, demonstrating that the combination of both corridors could provide safer extensive resection.

CASE PRESENTATION

History and examination

A 27-year-old Japanese woman presented with a headache was discovered incidentally to have a tumor mass 1 cm in diameter near the right superior orbital fissure [Figure 1a]. She had no family history or another clinical presentation of neurofibromatosis. A month after the tumor was found, the patient became pregnant. At the 38th gestational week, she developed ptosis, and magnetic resonance imaging (MRI) showed that the tumor had grown rapidly, extending into the orbit [Figure 1b]. The mass showed mixed intensity but mostly hyperintensity on T2 [Figure 1b]. Head computed tomography (CT) revealed an enlargement of the superior orbital fissure [Figure 1c]. We chose to delay the resection until after delivery. The patient subsequently suffered from right abducens palsy, mildly dilated pupil, and proptosis just before delivery [Figure 1d]. Two months after an uneventful spontaneous vaginal delivery at 41 gestational weeks, further imaging studies were conducted.

MRI showed that a contrast-enhanced mass with a clear boundary and diameter of 5 cm had extended into the right intraconal space of the orbit through the superior orbital fissure to displace the optic nerve, causing proptosis [Figure 1e].

Based on the symptoms[39] and MRI and CT findings, a suggestive diagnosis of schwannoma of the ophthalmic division of the right trigeminal nerve was made. Tumor resection by EETOA was then planned.

Combined EETOA

The operation involved a collaboration of neurosurgeons and otolaryngologists [Figure 2a and Video 1]. The patient was placed in the supine position with her head on a soft pad and was registered for electromagnetic field guide navigation (Stealth Station ENT®, Medtronic, Minneapolis, MN, USA). A 4K, two-dimensional, rigid, 0° endoscope (Olympus, Tokyo, Japan) was used for visualization. Under general anesthesia, the right endonasal endoscopic approach was performed to fully open the ethmoid sinus and expose the maxillary sinus and lamina papyracea. A bilateral rescue flap incision was made, and the sphenoid sinus was widely opened. The lamina papyracea was subsequently fractured out. The right anterior and posterior ethmoid arteries were coagulated and severed to facilitate access to the intraorbital region from the endonasal approach. The right sphenopalatine artery was coagulated and severed. The base of the pterygoid plate was drilled, and the bulge of the cavernous sinus content was exposed [Figure 2b]. The internal carotid artery (ICA) was identified by Doppler sonography, and the cavernous sinus wall lateral to the ICA was carefully incised. The exposed tumor was soft and removed piece by piece [Figure 2c]. The intraoperative pathological diagnosis confirmed the tumor as schwannoma. The tumor mass was subsequently removed in a subcapsular fashion. After removing the cavernous sinus portion, the periorbita was incised to expose the intraorbital region. However, the visibility and operative maneuverability were limited. Transorbital ultrasonography showed a residual mass in the intraorbital region; thus, we decided to perform the transorbital approach additionally. A supraorbital incision was made, and the periosteum was dissected to reach the superior orbital rim [Figure 2d]. The eyeball was intermittently depressed with a spatula, and the residual tumor was removed through this corridor piece by piece [Figure 2e]. The resection was continued until the removal cavity was continuous with the endonasal removal cavity. Subsequent intraoperative MRI showed gross total resection of the tumor [Figures 3a, 3b]. A collagen matrix overlaid the lateral part of the cavernous sinus, and the eyebrow incision was subcutaneously sutured with a 3-0 polydioxanone suture. A running 5-0 nylon suture was then made to close the skin. The entire procedure lasted 5.5 hours, including the time interval for performing intraoperative MRI.

Pathological examination showed spindle-shaped cell tumors with hyaline small blood vessels [Figure 3c]. Immunohistochemical staining showed that the tumor was positive for S-100 [Figure 3d], thereby confirming the intraoperative diagnosis of schwannoma.

Combining the intraoperative and pathological findings, the final diagnosis was trigeminal ophthalmic division schwannoma.

The patient recovered completely without any complications. She was discharged home 4 days postoperatively. At the 4-month follow-up, her abducens nerve palsy and proptosis had resolved completely. The mildly dilated pupil remained, possibly due to irreversible damage to the ciliary ganglion.

DISCUSSION

Since trigeminal schwannoma progresses to various sites and is anatomically complex, an understanding of surgical anatomy and selecting the appropriate approach

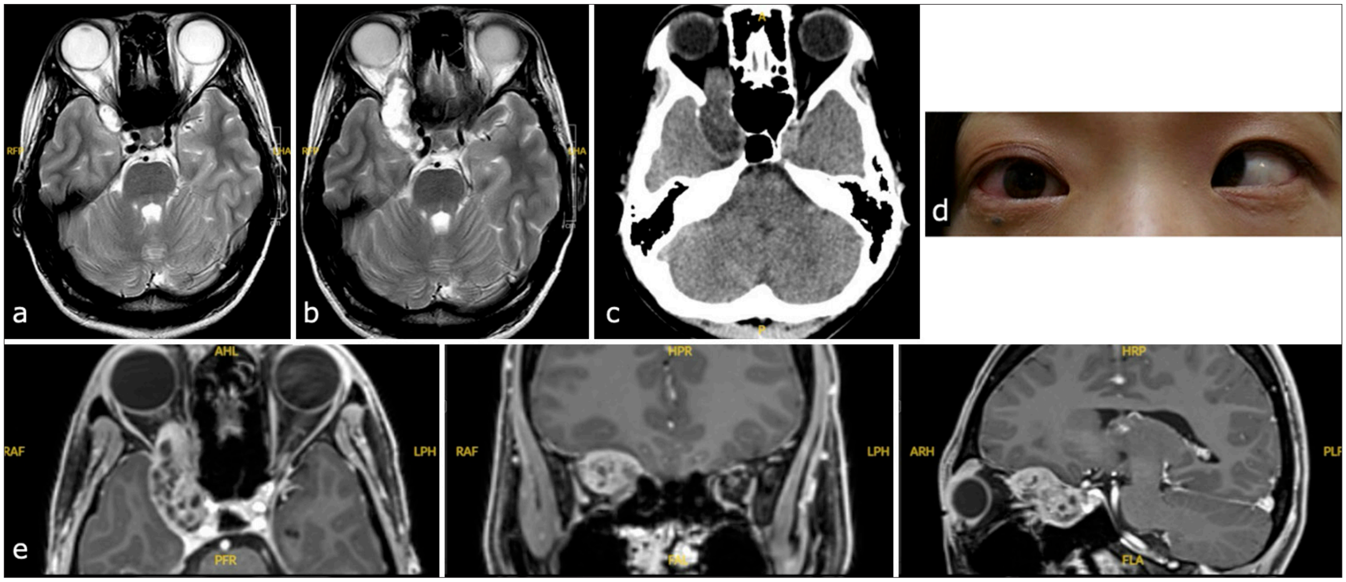

Figure 1: (a) Initial magnetic resonance T2-weighted imaging of the tumor. (b)The tumor grew rapidly during pregnancy and extended to the intraorbital area. (c) Head computed tomography showing the enlargement of superior orbital fissure. (d) At the last trimester, the patient developed right abducens palsy and a mildly dilated pupil. (e) Preoperative Gadolinium-enhanced T1 imaging of the tumor in axial, coronal, and sagittal views from left to right..

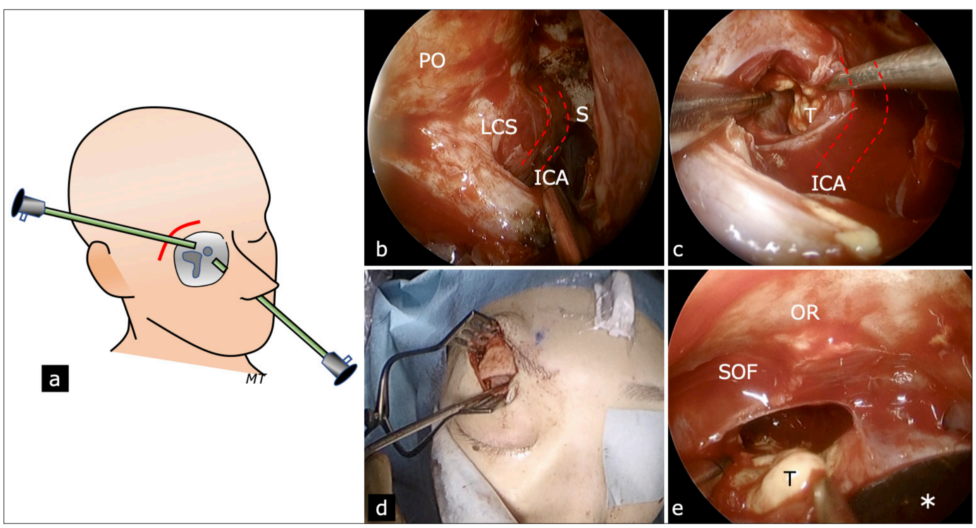

Figure 2: (a) Schema of multiport combined endoscopic endonasal and transorbital approach via an eyebrow incision. (b) After exposing the periorbita and lateral compartment of the cavernous sinus, the internal carotid artery was identified by Doppler sonography and electromagnetic field guide navigation. (c) The lateral cavernous sinus compartment was incised, and the tumor was removed in a subcapsular fashion. (d) An eyebrow incision was made, and the superior orbital rim was exposed in a subperiosteal fashion. After the orbit was compressed downward with a spatula (*), the endoscope was introduced, and the tumor was removed piece by piece. (e) An enlarged superior orbital fissure was noted. ICA:Internal carotid artery; LCS :Lateral compartment of the cavernous sinus; OR :Optic roof; PO :Periorbita; S:Sella; SOF:Superior orbital fissure; T:Tumor.

are important considerations. Yoshida and Kawase[40] proposed a classification that divides intracranial trigeminal schwannomas into three components: middle cranial fossa (M), posterior fossa (P), and extracranial fossa (E). Many complex-type tumors are classified as MP, ME, or MPE depending on the components involved in the progression of the trigeminal schwannoma. Fukaya *et al.*[16] analyzed the progression of trigeminal schwannoma based on this MPE classification and reported that the frequency was M: 14%; P: 21%; E: 7%; MP: 39%; ME: 12%; and MPE:

7%. The least frequent extracranial type can be further subdivided into orbital (E1 type), pterygopalatine fossa, and infratemporal fossa (E2 type). Trigeminal schwannomas extending into multiple fossae can be totally resected according to the following single-stage surgical techniques: MP by the anterior transpetrosal approach, ME by the zygomatic or orbitozygomatic infratemporal approach, and MPE by the zygomatic transpetrosal approach. The zygomatic infratemporal approach has also been used to resect type E2 schwannomas. Our case was predominantly an E1-type tumor with a lateral cavernous sinus compartment. Primary intraorbital schwannomas are rare, accounting for approximately 1–2% of orbital tumors.[23,31] Intraorbital schwannomas have also been associated with neurofibromatosis,[26] and the accelerated growth of intraorbital schwannoma during pregnancy is rare.[10,21,35] The underlying mechanism of the latter remains unclear, but it may be caused by hormonal influence and/or increased blood volume during pregnancy.[34]

Video 1: Combined endoscopic endonasal and transorbital approach.

Approaches to intraorbital tumors are broadly divided into the transcranial, endonasal, and transorbital approaches [Table 1]. Paluzzi *et al*.[28] proposed the simple "Round-the-Clock" surgical access algorithm, in which the location of the pathology within the orbit observed at the coronal section and relative to the optic nerve should dictate the choice of approach. The EEA is from 1 to 7 o'clock, the lateral approach from 8 to 10 o'clock, and the transcranial approach from 9 to 1 o'clock. In our case, the tumor was located in the 10–2 o'clock area, which indicated the transcranial approach rather than the endonasal approach.

The transcranial approach to the orbit[32] is the surgical method most familiar to neurosurgeons. The transcranial approach is most suitable for lesions located above the optic nerve at the medial and orbital tip of the optic nerve. This method is most appropriate for lesions that span the orbit, such as tumors with intracranial and intraorbital extensions. Schwannomas, including E1, arise from the first division of the trigeminal nerve (supraorbital and supratrochlear branches), which makes the transcranial approach the optimal method. It provides a wide exposure of the tumor and excellent surgical maneuverability, and the indication is not only for the intraorbital region but also for intracranial extensions. Combined with temporal bone and zygomatic resection, the transcranial approach can be performed to resect schwannomas involving multiple compartments.[16] However, the transcranial approach is disadvantageous in terms of invasiveness and postoperative cosmetics because it involves craniotomy and detachment of the temporal muscle.

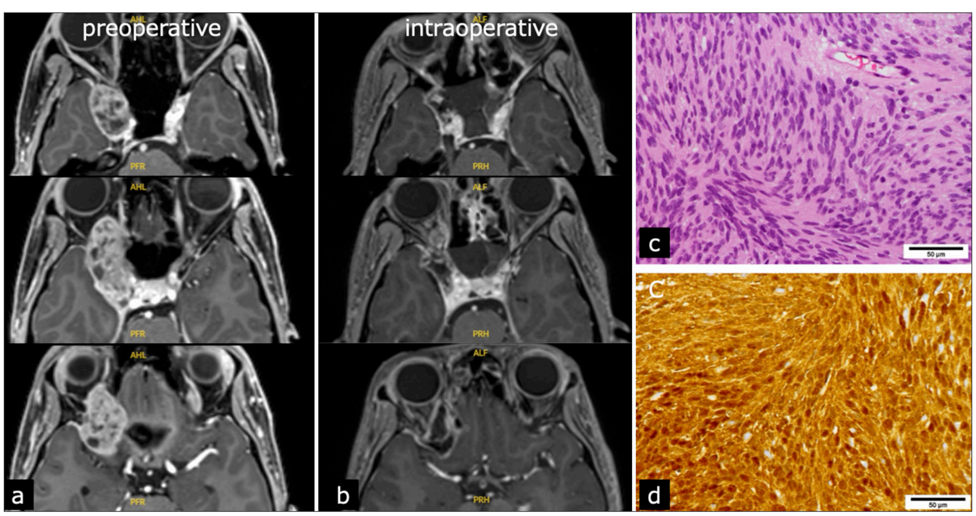

Figure 3: (a) Preoperative axial magnetic resonance images. (b) intraoperative axial magnetic resonance images demonstrating gross total tumor removal. (c)Hematoxylin and eosin staining showing spindle-shaped cell tumors. scale bar = 50um (d) Tumor cells are positive for S-100. scale bar = 50um.

Table 1: Features of various surgical approaches to the orbit.

	Indications[24]	Advantages	Disadvantages	Extensibility
Craniotomy	9–1 o'clock	Excellent surgical maneuverability; most familiar to neurosurgeons.	Invasive: long skin incision	Cavernous sinus, middle fossa, infratemporal fossa, posterior fossa
Lateral orbitotomy	8–10 o'clock is best suited for lesions lateral to the optic nerve.	Less invasive than craniotomy	Damage to the frontal branch of the facial nerve; limited visualization	Cavernous sinus, middle fossa, and superior orbital fissure, but the surgical field is limited and requires surgical experience
Endoscopic endonasal approach	1–7 o'clock	Excellent cosmetic results identification of the internal carotid artery is relatively easy.	Poor maneuverability; injury to ethmoidal arteries access to lesions lateral to the optic nerve is not possible.	Orbital decompression, optic nerve decompression infratemporal fossa, lateral cavernous sinus, Meckel cave (transpterygoid approach)
TONES approach	Depends on the lesion	The parallel axis to the orbit permits shorter access depending on the lesion	Requires eyeball retraction; unfamiliar anatomy	Access to the cavernous sinus and middle fossa is possible
EETOA	Depends on the lesion	Better maneuverability and visualization than a single approach only	Requires an experienced team; combining the two approaches avoids complications	Cavernous sinus, middle fossa

TONES: Transorbital neuroendoscopic surgery, EETOA: Endoscopic endonasal and transorbital approach

Lateral orbitotomy involves opening the outer rim of the orbit and entering the posterior surface of the eye from the lateral side.[24] Since a skin incision is made on the face, special consideration is required for wound treatment and injury to the facial nerve. Lateral orbitotomy is less invasive than a craniotomy, but the location of the intraorbital tumor that can be approached is mostly limited to the lateral part of the orbit (8–10 o'clock).[28] Lateral orbitotomy can also be performed for tumors in the orbital apex, superior orbital fissure, cavernous sinus, and middle cranial fossa by changing the orientation of the microscope or, in more experienced hands, combined with the endoscope.[6,38]

Cases employing the endonasal approach to resect orbital tumors are increasing.[8,33] The endonasal approach does not require an external incision, has excellent cosmetic results, and is less invasive than a craniotomy. It can also be useful for orbital or optic nerve decompression[3] because it provides excellent exposure of the optic canal and the orbital apex with minimal invasiveness. The lateral compartment of cavernous sinuses can be simultaneously approached endonasally,[19] but compared with the transcranial approach, maneuverability and exposure are limited. In recent years, the endonasal transpterygoid approach has been used in resecting pterygopalatine and infratemporal fossa lesions.[20]

The transorbital approach has undergone recent developments, and its indication has been extended by endoscopy.[1] Park *et al.*[29] conducted a retrospective multicenter analysis of 25 patients who underwent endoscopic surgical treatment for trigeminal schwannoma. Thirteen patients (52%) underwent the EEA, whereas 12 (48%) underwent the endoscopic transorbital approach. They demonstrated that the endoscopic transorbital approach provides adequate access and increases resectable tumor volume for trigeminal schwannomas located in the middle fossa or dumbbell-shaped tumors located in the middle and posterior fossae.

A multiport approach combining the endoscopic endonasal and endoscopic transorbital approaches has been recently reported. Ciporen *et al.*[11] have reported the usefulness of the transorbital endoscopic approach to the anterior middle skull base, which they called transorbital neuroendoscopic surgery (TONES). They combined TONES with the endonasal approach and showed that the addition of TONES was useful because of the shorter working distances, improved visualization, and excellent working angles that offered more direct access to the pituitary gland, suprasellar region, clivus, and medial and lateral cavernous sinus than the EEA alone. Dallan *et al.*[13] also reported their preliminary results for the application of the multiport EETOA approach in cadaver and clinical cases. The advantages of the combined approach are shorter access to the target, prevention of the crossing of nerve or major vessels, and multi-perspective visualization. In the case of temporal fossa invasion, lesions that are too posteriorly extended and major vessel involvement were not suitable for the multiport approach, and the transcranial approach should be considered. Lee *et al*.[25] described the approach combining EEA and TONES for various lesions, including two cases of schwannoma. EEA and TONES are connected at specific anatomical areas: the horizontal portion of the greater sphenoid wing, the foramen ovale, and the optic canal. A combined approach was thus concluded as a useful strategy for complex lesions involving multiple compartments of the skull base. The concept of connection areas was further investigated in a cadaveric study by Guizzardi *et al*.[17,18] At the anterior cranial fossa, the connection area was found at the sphenoid planum level; in the middle cranial fossa, it was at the Mullan triangle; and in the posterior cranial fossa, the connection area was just behind the medial portion of the petrous apex. Bhuskute *et al*.[4,5] reported the advantages of the multiport endoscopic endonasal and medial transorbital approach[12,27,30] to the contralateral lateral recess of the sphenoid sinus, petrous apex, and retrocarotid area compared to the endonasal approach with the contralateral transmaxillary approach. The multiport endoscopic endonasal and medial transorbital approach offer the advantage of preserving the contents of the pterygopalatine fossa and the vidian nerve, which are frequently sacrificed during a conventional transpterygoid approach. This technique provides superior visualization and better instrument maneuverability.

Our patient had a large lesion extending from the cavernous sinus to the orbit, which ostensibly indicated the transcranial approach. However, in the pursuit of a minimally invasive method and shorter hospitalization, we decided to employ endoscopy in combination with both the nasal and transorbital approaches. We inferred that the intraorbital region could not be removed through the endonasal approach alone. Furthermore, removal of the cavernous sinus region would probably be difficult through the transorbital approach alone. Furthermore, decompressing the orbit and managing the ICA would have been particularly difficult.

In the case of EETOA surgery, a part of the orbital paper-like plate was first separated into an elliptical shape through the nasal approach to provide mobility. Pressure on the eyeball during the transorbital operation was relieved, and sufficient working space was secured to perform the intraorbital operation more safely. Although abducens nerve monitoring was not performed in this case, a safer surgery could have been possibly performed if monitoring was performed at the time of incision. In addition, we considered the possibility of additionally performing supraorbital craniotomy if transorbital manipulation was insufficient and made the tumor difficult to resect. The eyebrow incision is more familiar to neurosurgeons[36] than the superior crease incision and provides a possible extension to craniotomy if needed.

CONCLUSION

This report demonstrated that EETOA was successfully performed in the resection of an intraorbital schwannoma. The findings highlight how surgeons may apply multiple corridors in successfully accessing and resecting complex skull base tumors.

Ethical approval

The research/study was approved by the Institutional Review Board at Kyoto University Hospital, number R2088, dated February 01, 2020.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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