CASE REPORT

Supraorbital nerve schwannoma in a young Chinese man: a case report and review of the literature

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Abstract

Schwannomas of peripheral branches of the trigeminal nerve are rare. We report a 23-year-old Chinese man who underwent anterior orbitotomy through an upper eyelid crease incision approach for resection of a supraorbital nerve schwannoma. Clinical features, imaging findings, and management considerations are discussed, and the literature reviewed.

Key words: *Eyelids; Neurilemmoma; Orbit*

Introduction

Schwannomas are benign, slow-growing tumors arising from the myelin sheath Schwann cells of the peripheral nerves. Orbital schwannomas are uncommon and account for 1% to 2% of all orbital tumors.[1] They usually originate from the ophthalmic division of the sensory branch of the trigeminal nerve, most commonly the supraorbital and supratrochlear nerves.[2] We report an anterior orbitotomy through an upper eyelid crease incision approach for resection of a supraorbital nerve schwannoma.

Case Presentation

In February 2015, a 23-year-old man with a history of recurrent craniopharyngioma and persistent visual impairment presented with a painless mass in the superomedial aspect of the left upper eyelid. He had no diplopia, ophthalmoplegia, proptosis or chemosis, or any deterioration in visual acuity. Confrontation testing revealed a left medial inferior quadrantanopia, which had been documented in 2008 as an old defect with optic atrophy, consistent with left optic disc pallor on fundi visualization. Ophthalmologic examination was unremarkable. Systemic evaluation excluded neurofibromatosis.

Contrast-enhanced computed tomography of the orbit demonstrated a well-circumscribed, homogenous, moderately enhancing ovoid solid mass of 1.2 cm × 0.8 cm × 0.8 cm at the superomedial aspect of the extraconal left orbit, anterosuperior to and abutting the globe. There were no aggressive features (bony destruction or invasion to surrounding structures), fluid, macroscopic fat component, or evidence of calcification. The left globe, lacrimal gland, and extraocular muscles were normal (**Figure 1**). Contrast-enhanced magnetic resonance imaging (MRI) showed a well-defined, homogenous, T1-hypointense and T2-hyperintense, avidly enhancing lesion of 1.1 cm × 1.0 cm × 1.0 cm, with no calcification or soft tissue or osseous invasion. There were no internal T1-hyperintense signals or fluid levels (**Figure 2**). The lesion had been identified on MRI in 2014. Owing to the static nature of the lesion, preliminary diagnoses included cavernous hemangioma and nerve sheath tumors.

In 2000, the patient had undergone craniotomy and excision of a craniopharyngioma. He had complications of panhypopituitarism and mild renal impairment but remained free of recurrence until 2015 when MRI of the pituitary gland incidentally found a well-circumscribed, T1-hyperintense mass of 1.7 cm × 1.9 cm × 1.8 cm at the pituitary fossa. A diagnosis of recurrent craniopharyngioma was made.

In January 2016, the patient underwent concomitant removal

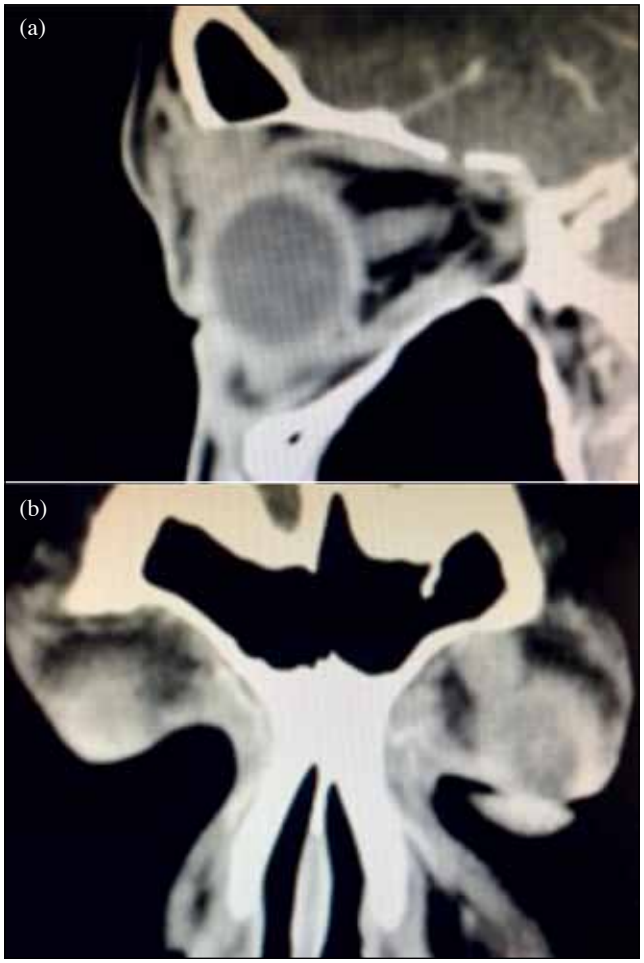

Figure 1. (a) Sagittal and (b) coronal views of contrast-enhanced computed tomography of the left orbit showing a moderately enhancing ovoid solid mass at the superomedial aspect of the extraconal left orbit. There are no aggressive features (bony destruction or invasion to surrounding structures), fluid, macroscopic fat component, or evidence of calcification. The left globe, lacrimal gland, and extraocular muscles are normal.

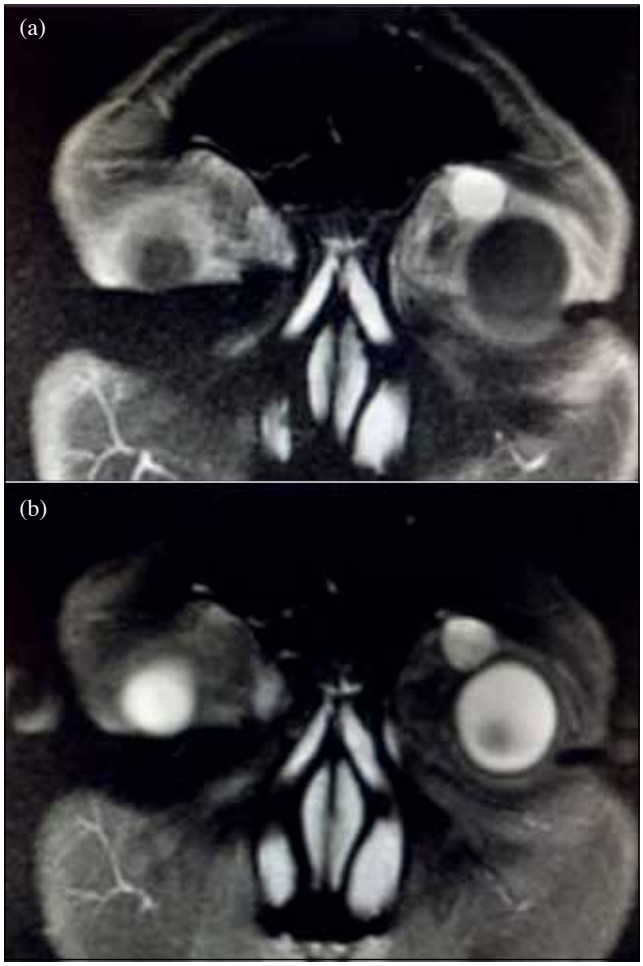

Figure 2. (a) T1-weighted and (b) T2-weighted contrast-enhanced magnetic resonance imaging of the orbits showing a T1-hypointense and T2-hyperintense, avidly enhancing lesion, with no calcification or soft tissue or osseous invasion. There are no internal T1-hyperintense signals or fluid levels.

of the pituitary and orbital masses in the same setting. For the orbital mass, anterior orbitotomy without bone flap through an upper eyelid crease incision was performed. Intra-operatively, the mass was noted to be arising from the supraorbital nerve. The resected oval mass was smooth and measured 1.2 cm × 1.0 cm × 1.1 cm, with a pale-yellow cut surface. Microscopic examination showed a circumscribed proliferation of spindle cells arranged in fascicles with nuclear palisading around fibrillary processes, consistent with Antoni A areas with Verocay bodies. The spindle cells contained elongated, wavy nuclei with tapered ends. There was no cytological atypia (**Figure 3a**). Immunohistochemical staining with S100 demonstrated diffuse positivity of the spindle cells for S100, establishing the diagnosis of schwannoma of the left supraorbital nerve (**Figure 3b**). Postoperatively, the patient reported mild sensory deficits over the distribution of the ophthalmic division of the left trigeminal nerve, which resolved after 6 months. Recovery was uneventful at 1-year follow-up.

Discussion

Supraorbital nerve schwannomas are rare and only 10 cases have been reported. Orbital schwannomas primarily affect individuals aged 20 to 40 years.[3] Approximately 10% to 15% of orbital schwannomas are associated with neurofibromatosis and this type has an earlier onset, compared with the isolated type. The age of onset of our patient was 23 years, which is relatively early, given no personal or family history of neurofibromatosis.

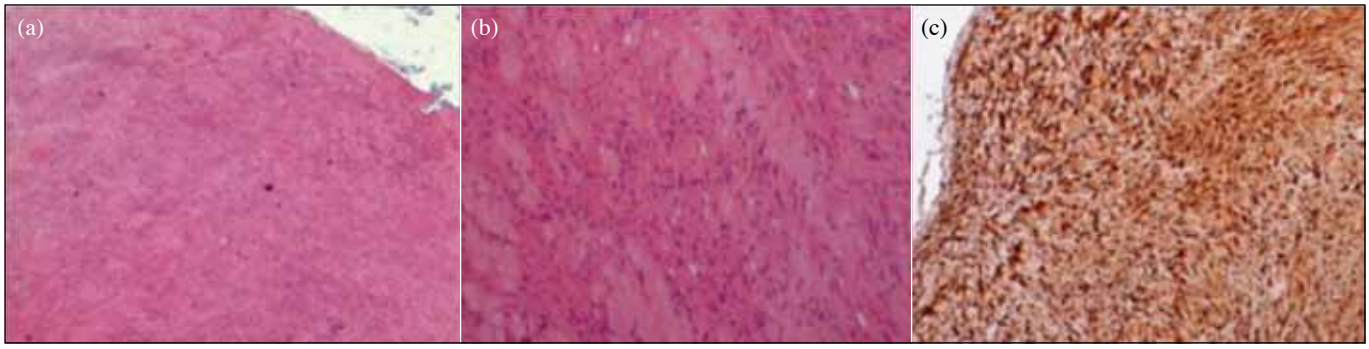

Figure 3. (a and b) Proliferation of spindle cells arranged in fascicles with nuclear palisading around fibrillary processes, consistent with Antoni A areas with Verocay bodies. The spindle cells contain elongated, wavy nuclei with tapered ends. There is no cytological atypia (hematoxylin and eosin, 40× and 100×, respectively). (c) Diffuse positivity of the spindle cells for S100 confirms the diagnosis of schwannoma (immunohistochemical staining with S100, 40×).

Table. 66 cases of orbital schwannomas reported in the literature[7-58]

Study	Sex/ age, y	History of neuro-fibromatosis	Tissue of origin / special feature	Location	Surgical approach	Treatment outcome
Skeoch,[7] 1956	M/41	-	-	-	-	Uneventful recovery
Horie et al.[8] 1982	F/41	-	Supraorbital nerve	-	Subfrontal orbitotomy	Uneventful recovery
Shields et al.[9] 1986	M/33	No	-	-	Lateral orbitotomy	Uneventful recovery
Grover et al.[10] 1993	F/45	-	-	Intraconal	Lateral orbitotomy	Uneventful recovery
Lam et al.[11] 1997	F/54	-	Ophthalmic nerve	Extraconal	Sub-brow incision	Uneventful recovery
Maesawa et al.[12] 1998	F/40	-	Ophthalmic nerve	-	Fronto-orbitozygomatic craniotomy	Residual sensory symptoms
Khwarg et al.[13] 1999	F/52	No	-	Intraconal	Lateral orbitotomy via an eyelid crease incision	Uneventful recovery
Tsuzuki et al.[14] 2000	F/62	No	Oculomotor nerve	Intraconal	Microsurgical lateral approach	Uneventful recovery
Goel et al.[15] 2003	M/18	No	Cavernous sinus	Extraconal	Lateral orbitotomy	Uneventful recovery
Schick et al.[16] 2003	M/36	Personal: type 2	-	Intraconal	Extradural pterional approach	Uneventful recovery
	F/24	Personal: type 1	-	Extraconal	Supraorbital approach	Resolution of proptosis but visual acuity remained poor
	F/43	-	-	Intraconal	Inferior-medial transconjunctival approach	Uneventful recovery
	M/50	No	Supraorbital nerve	Extraconal	Supraorbital approach	Uneventful recovery
	F/60	-	-	Intraconal and extraconal	Extradural pterional approach	Development of amaurosis with ptosis
Chang et al.[17] 2003	F/38	No	-	Intraconal	Lateral orbitotomy	Uneventful recovery
Barbagallo et al.[18] 2004	M/65	No	Supraorbital nerve	Extraconal	Fronto-orbitozygomatic craniotomy	Uneventful recovery
El-Bahy[19] 2004	M/40	No	Spheno-orbital	Extraconal	-	-
Subramanian et al.[20] 2005	F/18	-	- / With cystic degeneration	Intraconal	Combined medial and lateral orbitotomy	Resolution of proptosis but deterioration in vision
	M/40	-	- / With cystic degeneration	Intraconal	Lateral orbitotomy	Uneventful recovery
	F/78	-	- / With cystic degeneration	Intraconal	Enucleation	-
	M/48	-	- / With cystic degeneration	Intraconal	Lateral orbitotomy	Uneventful recovery
Tezer et al.[21] 2006	F/16	-	Infraorbital nerve	-	Subciliary incision	Residual sensory symptoms
Furuno et al.[22] 2006	F/74	-	Oculomotor nerve	Intraconal	Transcranial approach	Uneventful recovery
Boustred[23] 2007	F/5	Personal	Supraorbital nerve	-	Endoscopic approach	Uneventful recovery
Sales-Sanz et al.[24] 2007	F/49	No	- / Bilateral synchronous schwannomas	Extraconal	Subciliary inferior orbitotomy	Uneventful recovery
Colapinto et al.[25] 2007	M/68	No	Inferior oblique	Intraconal	Anterior orbitotomy	Uneventful recovery
Sugo et al.[26] 2007	F/34	No	-	Intraconal	Orbital unroofing by a frontotemporal approach	Uneventful recovery
Tanriover et al.[27] 2007	F/34	No	Oculomotor nerve	Extraconal	Pterional approach	Uneventful recovery

Table. (cont'd)

Shamim et al.[28] 2008	F/11	No	Oculomotor nerve	Intraconal	-	Uneventful recovery
Irace et al.[29] 2008	M/55	No	Abducens nerve	Intraconal	Modified microsurgical lateral orbitotomy	Uneventful recovery
Garg et al.[30] 2008	F/35	-	Infraorbital nerve	Extraconal	Inferior orbitotomy via an eyelid crease incision	Uneventful recovery
Youens et al.[31] 2008	F/51	No	- / Hybrid type	Extraconal	Anterior orbitotomy via an eyelid crease incision	Uneventful recovery
Kiratli et al.[32] 2008	F/12	Personal and family	Lateral rectus	Intraconal	Fractionated stereotactic radiotherapy	Uneventful recovery
Miliaras et al.[33] 2008	M/62	No	- / Malignant type	Intraconal	Anterior orbitotomy with radiotherapy	Intracranial recurrence, patient died 13 months after diagnosis
Karkas et al.[34] 2008	M/14	No	Infraorbital nerve	Extraconal	osteoplastic maxillotomy approach	Uneventful recovery
de Silva et al.[35] 2009	F/66	No	Lacrimal gland	Extraconal	Lateral orbitotomy	Uneventful recovery
	F/30					
Hironaka et al.[36] 2010	M/58	No	Ophthalmic nerve	Intraconal	Fronto-orbitozygomatic craniotomy	Residual sensory symptoms
de Jong et al.[37] 2010	M/44	No	Supraorbital nerve / ancient type	-	Superolateral orbitotomy	-
Birkholz et al.[38] 2010	F/23	No	-	Intraconal	Stereotactic radiosurgery	-
Hayashi et al.[39] 2010	F/46, F/46, F/46, M/46	No	Cavernous sinus	Extraconal	Gamma knife surgery	Uneventful recovery for tumor that was intracavernous; cases with dumbbell lesions exhibited visual deterioration
Brucoli et al.[40] 2011	F/75	No	-	Extraconal	Lateral orbitotomy	Uneventful recovery
Gunduz et al.[41] 2011	M/40	No	-	Intraconal	Orbitotomy via a superonasal skin approach	Uneventful recovery
Pushker et al.[42] 2011	F/62	No	-	Extraconal	Anterior orbitotomy	-
Denadai et al.[43] 2012	F/35	No	Supraorbital nerve	-	Supraorbital orbitotomy	Uneventful recovery
Izumo et al.[44] 2012	M/67	-	Supraorbital nerve	Extraconal	Anterior orbitotomy	Residual numbness
Nagashima et al.[45] 2012	M/5	No	Oculomotor nerve	Intraconal	Frontotemporal craniotomy	
Rato et al.[46] 2012	M/42	No	Abducens nerve	Intraconal	Lateral orbitotomy	Uneventful recovery
Kron et al.[47] 2012	M/59	No	- / Recurrence, possible schwannomatosis	-	Anterior orbitotomy	Diplopia and ptosis requiring surgical treatment
	F/5	No	- / Recurrence	-	Anterior orbitotomy	
Feichtinger et al.[48] 2013	F/53	-	Abducens nerve	Intraconal	Lateral orbitotomy	Uneventful recovery but with some permanent deficits
Bassily et al.[49] 2013	M/23	-	Ethmoidal air cell	Extraconal	Anterior orbitotomy via an eyelid crease incision	Uneventful recovery
Claros et al.[50] 2014	F/29	No	-	Intraconal	Lateral orbitotomy	Uneventful recovery
Mortuza et al.[51] 2014	M/56	No	Sclera / ancient type	Intraconal	Enucleation	-
Chacko et al.[52] 2014	F/66	-	Supraorbital nerve	-	-	Uneventful recovery
Comez and Muratli[53] 2014	F/65	No	-	Extraconal	Subciliary incision	Uneventful recovery
Koktekir et al.[54] 2014	F/41	No	Opthalmic, maxillary and supratrochlear nerves	Intraconal	Lateral orbitotomy	Recurrence a year later, completely resected. Unremarkable since
	F/29	No	Ophthalmic nerve	Intraconal	Anterior orbitotomy via an eyelid crease incision	Uneventful recovery
	M/57	No	-	Intraconal and extraconal	Anterior orbitotomy via an eyelid crease incision	Uneventful recovery
Taubenslag et al.[55] 2015	M/31	No	Supraorbital nerve / hybrid type	Extraconal	Anterior orbitotomy via an eyelid crease incision	-
Li et al.[56] 2015	M/27	No	Superior oblique muscle	Intraconal	Anterior orbitotomy	Uneventful recovery
Jacquesson et al.[57] 2015	M/49	-	Maxillary nerve	Extraconal	Expanded endoscopic endonasal approach	Uneventful recovery
Gupta et al.[58] 2017	F/39	No	- / Concomitant orbital cavernous hemangioma and schwannoma	Intraconal	Inferior transconjunctival orbitotomy	Uneventful recovery

Orbital schwannomas commonly present with a painless proptosis. Rarely, when the supraorbital nerve is affected, schwannomas may cause numbness and/or pain in the distribution of the trigeminal nerve, mimicking symptoms of sinusitis.[4] On MRI, the tumor is classically hypointense on T1-weighted and hyperintense on T2-weighted images, with avid post-gadolinium enhancement.[5] Histological examination demonstrates Antoni A or Antoni B areas, Verocay bodies, and positivity for S-100 immunohistochemical stain.

The surgical approach varies with the location and extent of the tumor as well as the nerve involved. The fronto-orbitozygomatic craniotomy allows wider access and exposure of orbital surfaces and content, and has been used for the removal of large intraorbital tumors. In our patient, anterior orbitotomy through an upper eyelid crease incision was used without a bone flap. This allows dissection of comparatively avascular tissue planes and minimizes transection of the orbicularis muscle and lymphatic channels, resulting in negligible scarring postoperatively.[6]

The literature was reviewed by searching the PubMed using keywords: 'orbital' and 'schwannoma'. In 32 cases, the diagnosis was not orbital schwannoma or information was inadequate. 66 cases in 27 men and 39 women were orbital schwannoma (**Table**).[7-58] The overall median age of onset was 41 years (41 years in women, 43 years in men). Only four cases had personal or family history of neurofibromatosis; 19 cases had unknown medical history. The median age of onset was earlier in those with personal or family history of neurofibromatosis than in those with isolated orbital schwannomas (18 vs 43 years). The orbital schwannomas were intraconal in 29 cases, extraconal in 21 cases, and unknown in the rest. In only 10 cases (including our patient) did the schwannoma arise from the supraorbital nerve. The eyelid crease incision approach has only been used six times, and only once for the resection of a supraorbital nerve schwannoma.[55]

Conclusion

Early diagnosis and accurate dimension determination of orbital schwannoma is essential for complete removal, preservation of normal anatomy, and reinstitution of pre-morbid visual acuity and ocular movements.

Declaration

The authors have no conflicts of interest to disclose.

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