ORIGINAL RESEARCH

Orbital schwannoma: report of a case and review of the literature

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Acknowledgment
The authors would like to thank Joan S. K. Ng, FRCS, for her kind contributions in manuscript preparation.

Abstract

Orbital schwannoma is a rare orbital tumor comprising only about 1% of all orbital tumors. A 64-year-old Chinese male presented to our clinic with a 1-month history of unilateral proptosis, seeing double, and progressive painless visual loss. The proptosis was axial in nature, and an intraconal mass was revealed by an orbital computed tomographic scan. The mass was excised *en bloc* through a lateral orbitotomy approach. A histopathologic examination showed a well-encapsulated tumor composed of spindle cells arranged in both Antoni A and B patterns, which was compatible with the diagnosis of schwannoma. The differential diagnoses of intraconal orbital mass and the radiological and pathological findings of the lesion are described and discussed. A literature review of 60 published cases of orbital schwannoma is also presented.

Key words: Orbit, Schwannoma, Proptosis, Intraconal

Introduction

Orbital schwannoma is a rare, benign, encapsulated, peripheral nerve sheath tumor, usually arising from sensory branches of the trigeminal nerve.[1] It typically develops in middle-aged adults and produces a slowly evolving, well-tolerated proptosis.[2] There is no sex predilection.[3] Most cases are unilateral and solitary, although a bilateral case has been reported.[4] Malignant transformation of benign orbital schwannoma is rare, and this usually occurs in patients with neurofibromatosis.[5]

Schwannoma of the orbit grows very slowly and possibly intermittently and has no distinct clinical features.[6] Neglected tumors may attain a phenomenal size, causing exophthalmos, limitation of ocular movements, and blurring of vision through pressure effect on the optic nerve.[6]

The following is a clinicopathologic case report of a large intraconal orbital schwannoma causing significant impairment in vision and ocular motility.

Case report

A 64-year-old Chinese monk with no prior ocular problems presented to our clinic with a 1-month history of proptosis, seeing double, and progressively reduced vision in the left eye. There were no associated symptoms of ocular pain, headache, or systemic upset. Apart from a history of mild, well-controlled, non-insulin-dependent diabetes mellitus, he had enjoyed good health. Both trauma history and symptoms of thyrotoxicosis were negative.

His best-corrected visual acuity was 20/30 in the right eye, and 20/80 in the left eye. An afferent pupillary defect of the left eye was detected. An axial proptosis of 3 mm of the left eye was documented by Hertel exophthalmometry. Although no obvious strabismus could be demonstrated, ocular motility was markedly impaired in all directions, especially in vertical gaze. The orbital margins were well defined. There was no palpable mass, but increased

resistance to retropulsion of the left eye was noted. The anterior segment and intraocular pressure were normal. A fundal examination by indirect ophthalmoscopy showed bilateral, mild, nonproliferative diabetic retinopathy and a swollen, congested, hyperemic left disc with some choroidal folds.

A systemic examination revealed no enlarged lymph nodes as well as no sign of thyrotoxicosis and neurofibromatosis. The patient's chest was clear, and no abdominal mass was palpable.

Ultrasonography, using both A and B modes, of the left eye demonstrated a well-circumscribed retrobulbar mass with low internal reflectivity and fairly good sound transmission (**Figure 1**). An orbital computed tomographic (CT) scan showed a large, discrete, globular mass that was similar in size to the eye globe. It was situated intraconally, impinging on the posterior surface of the left eyeball and extending toward the optic canal. The optic nerve was inferiorly and medially displaced. There was no clear plane of demarcation between the mass and the optic nerve (**Figure 2**). Peripheral rim enhancement of the mass was seen in the contrast study. No calcification was demonstrated. The orbital walls were intact.

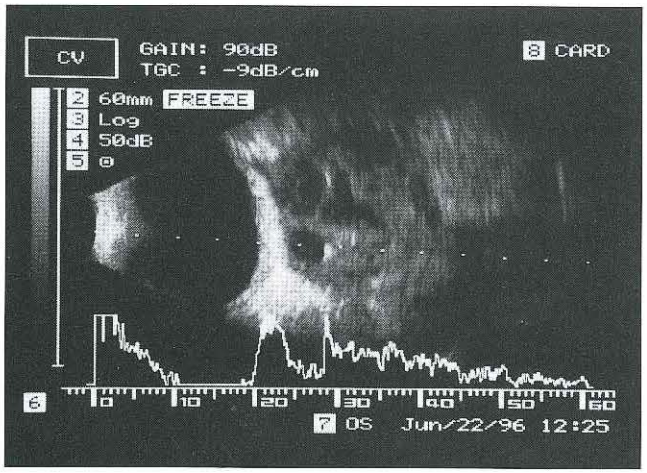

Figure 1. Ultrasonography of the left orbit showing a retrobulbar mass with multiple internal cystic spaces.

Because of the presence of optic nerve compression and the risk of malignancy, the patient elected to have the mass excised surgically. Lateral orbitotomy was performed while the patient was under general anesthesia. After the muscle cone was opened, a well-encapsulated brown-yellow mass was identified. It was located beneath the superior rectus muscle and surrounded the optic nerve superomedially. Although the tumor was in close contact with the optic nerve, it was removed en bloc successfully. It measured 2.5 x 2.0 x 1.5 cm in size. The eyeball was intact.

Nine months after the operation, the patient's visual acuity returned to 20/600. A pale optic disc was found in the left eye. Ocular motility returned to normal, and the patient had neither exophthalmos nor diplopia.

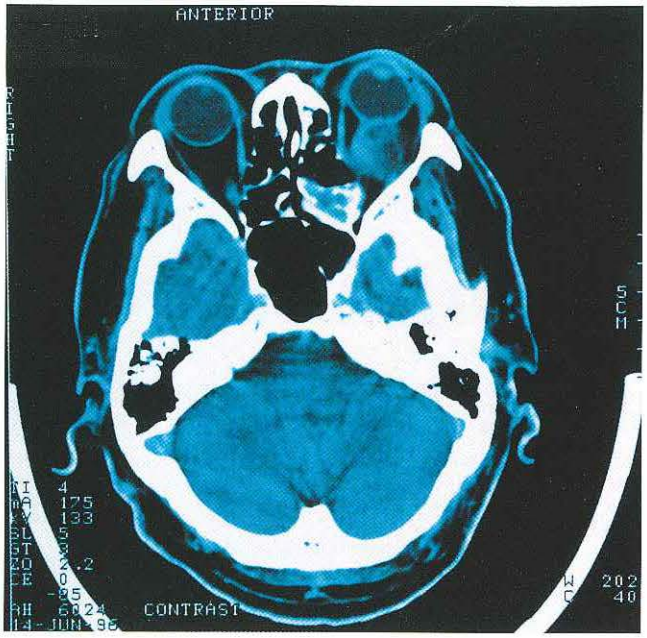

Figure 2. Computed tomogram of the orbits demonstrating a left well-defined intraconal mass.

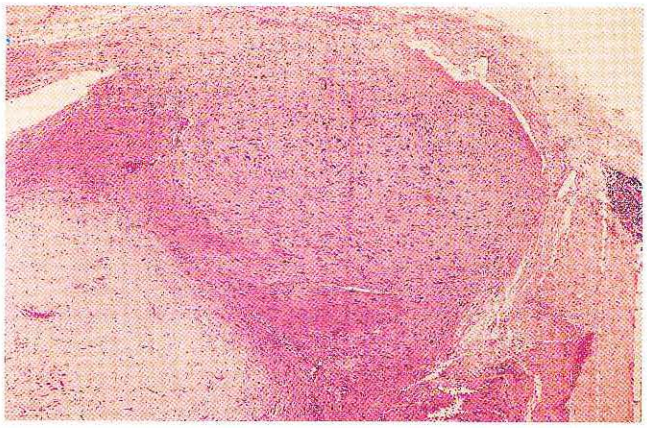

Figure 3. The tumor consists of spindle cells arranged in alternating Antoni A (middle and right side of field) and Antoni B (left lower side of field) patterns. (Hematoxylin-eosin, x 100)

Pathologic findings

The tumor was a globular mass of friable pinkish yellow tissue, measuring 2.5 cm in the greatest diameter. The capsule was firm and smooth. The cut surface showed necrotic hemorrhagic tissue with cystic spaces containing brownish yellow fluid. Sections showed a well-defined tumor consisting of 2 distinct alternating patterns (**Figure 3**). In some areas, there was predominance of benign, nonpigmented spindle cells with bland dark nuclei, indistinct cell membranes, and abundant intercellular basement membrane. This was typical of an Antoni A pattern of schwannoma (**Figure 3**). In other areas, there were large, haphazardly arranged spindle cells distributed in a myxomatous matrix. This is consistent with an Antoni B pattern of schwannoma (**Figure 3**). There were also large

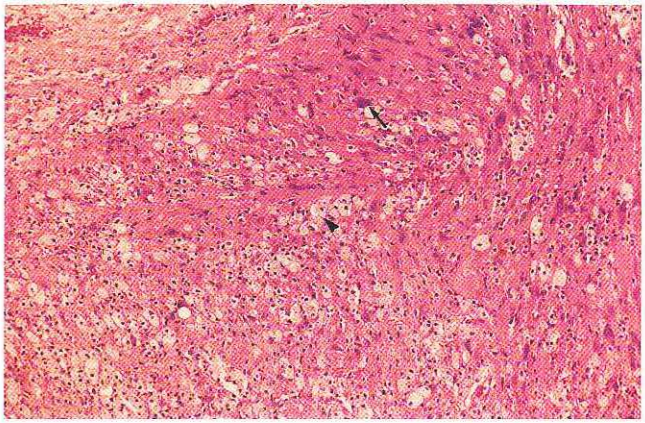

Figure 4. Higher magnification of an Antoni A area shows compact spindle cells, some of which show larger atypical nuclei (arrow). There are many foamy histiocytes (arrowhead) in the tumor. (Hematoxylin-eosin, x 400)

areas of hemorrhage, thrombosed vessels, and sheets of foam cell collections; focal areas showed degeneration atypical of tumor cells (**Figure 4**). The pathological diagnosis was schwannoma.

Discussion

Orbital schwannomas arise from the Schwann cells of peripheral nerves.[7] These tumors comprise approximately 1% of all orbital tumors, and are found in 1.5% of patients with neurofibromatosis type 1.[8] To further study the clinical characteristics of this uncommon tumor, we performed a literature search and reviewed 60 individually reported cases of orbital schwannoma between the period of 1925 and 1997 (**Table 1**). The age of presentation ranges from 15 months[9] to 72 years[3] (mean, 34.7 years). The male-female ratio is 26:34. The interval between the onset of symptoms and time of medical treatment ranged from 10 days[10] to 36 years.[1] This may determine the size of the tumors on presentation. The smallest tumor documented was 1 cm in diameter, while the largest one occupied the whole orbital cavity.[1]

Ocular complaints depend on the location and size of the tumors. The most common presenting symptom is exophthalmos, which occurred in 52 cases (86.7%), followed by visual disturbance (23 cases, 38%), limitation of ocular movement with diplopia (20 cases, 33.3%), ocular pain or headache (7 cases, 11.7%), and palpable mass around the periorbital area (4 cases, 6.7%).

In the present case, the patient had axial proptosis and progressive visual loss. The clinical impression of intraconal mass was confirmed by radioimaging. The initial clinical differential diagnosis included cavernous hemangioma, intrinsic optic nerve lesions like meningioma and glioma, orbital lymphoma, orbital pseudotumor, and metastatic tumors. Although orbital malignancies can sometimes produce a surprisingly circumscribed appearance on imaging, the relatively asymptomatic clinical course with the absence of pain and systematic upset made this diagnosis unlikely. Lymphoid tumor usually replaces the orbital fat in an irregular fashion rather than creating rounded masses on CT scan.[11] Orbital pseudotumor tends to extend into and obscure the orbital apex instead of having a well-demarcated posterior margin.[1] Magnetic resonance imaging (MRI) may help in differentiating a primary optic nerve tumor from a juxtaoptic nerve tumor and assessing the extent of the lesion. However, it was not done in our case because the echographic features of the mass were not characteristic of an intrinsic optic nerve tumor. Moreover, urgent operation was needed to relieve the optic nerve compression. With a presentation of painless axial proptosis and CT scan findings of a well-demarcated intraconal mass, cavernous hemangioma was the most likely diagnosis. However, the absence of multiple high internal echoes on the A-scan was atypical of cavernous hemangioma.

Although imaging studies are useful in the diagnosis of orbital schwannoma, there is no single pathognomonic diagnostic feature among these investigations. The diagnosis still depends on the histopathologic characteristics of an excisional biopsy specimen, which were characteristic of schwannoma in our specimen. On retrospective study, our case presented with the common features of orbital schwannoma, including proptosis and restricted ocular motility. The CT scan and ultrasound findings were also suggestive of orbital schwannoma. However, the short duration of symptoms and the rapid deterioration of vision were quite atypical to the diagnosis of orbital schwannoma. One possible explanation is that the tumor expanded very slowly, producing a well-tolerated proptosis without any pain. This slow progression of a relatively asymptomatic condition renders the patient less alert to seek medical attention. However, the tumor was located in the intraconal space, which has an intimate relationship with the optic nerve. Because of the limited intraconal space, when the tumor increased to a critical size, it directly compressed the optic nerve and resulted in rapid visual deterioration.

It is both difficult and challenging to identify the nerve of origin of the orbital schwannoma, particularly in cases with a mass of significant size. In the 60 cases reviewed, 23 (38.3%) were located within the muscle cone, 34 (56.7%) were located outside the muscle cone, and 3 (5.0%) were not mentioned in the reports. The nerves of origin were defined in 18 cases (30%). The ciliary nerve is the most common site of origin (7 cases), followed by the supraorbital nerve (5 cases) and the infraorbital nerve (2 cases). Other sites of origin include the superior maxillary nerve,[12] the superior branch of the oculomotor nerve,[1] the supratrochlear nerve,[13] and even the medial rectus muscle.[14] In the present case, the tumor most likely originated from the short posterior ciliary nerves adjacent to the optic nerve. Its intimate juxtaoptic nerve locus accounted for the difficulty of distinguishing it from the optic nerve on a CT scan.

Because orbital schwannomas are typically well encapsulated, the recommended treatment is complete surgical removal.[1] Recurrence is rare.[15] The surgical approach is dictated by the size and location of the lesion shown on axial and coronal CT scans and MRI. Most authors in our literature survey favor a lateral orbitotomy (Kronlein operation) approach. Depending on the extent and location

Table 1. Survey of 60 published cases of orbital schwannoma.

*Indicates that the information was not mentioned in this article.

Patient No.	Year of publication	Authors	Sex	Age/y	Presenting symptoms	Duration of symptoms	Presumed origin	Size/cm	Treatment	Follow-up and comments
1	1925	Cohen[16]	M	11	Right proptosis Restricted motility	2 years	Posterior ciliary nerve Intraconal	3.5 x 2.2	Lateral orbitotomy	Regained motility
2	1935	Sitchevska[17]	F	28	Right proptosis Blurred vision	2.5 years	Extraconal	2.0 x 3.0	Lateral orbitotomy	Slight enophthalmos after 9 months
3	1950	Standal[18]	M	8	Proptosis Left upper eyelid swelling	3 months	Extraconal	*	Irradiation	Malignant tumor Died 9 months after irradiation
4	1950	Standal[18]	M	1.5	Left upper eyelid swelling	1 month	Extraconal	*	Anterior orbitotomy	Malignant neurinoma with adjacent infiltration
5	1950	Standal[18]	F	4	Left proptosis Restricted motility	6 months	Extraconal	*	Lateral orbitotomy	No recurrence after 7 months
6	1950	Standal[18]	M	14	Right proptosis Blurred vision	4 months	Extraconal	Walnut-sized	Lateral orbitotomy	No recurrence after 4 months
7	1950	Standal[18]	F	20	Left proptosis Diplopia	2.5 years	Extraconal	*	Biopsy	Destruction of paranasal sinuses and anterior cranial fossa
8	1950	Standal[18]	M	33	Right proptosis Diplopia	4 months	Extraconal	*	Partial removal Irradiation	Destruction of right orbit
9	1950	Standal[18]	F	31	Left proptosis Blurred vision Squint	2 years	Intraconal	Walnut-sized	Piecemeal removal Irradiation	No recurrence after 10 years
10	1950	Standal[18]	F	23	Left proptosis Optic atrophy	*	Intraconal	Walnut-sized	Irradiation Excision	No recurrence after 8 months
11	1952	Shapiro and Scheffler[19]	F	24	Left proptosis Visual loss	Several months	Ciliary nerve Intraconal	*	Piecemeal removal	Uneventful
12	1956	Skeoch[20]	M	41	Left painless swelling	*	Extraconal	*	Excision	Uneventful
13	1961	Nissan[21]	F	18	Left proptosis Visual loss	2 years	Supraorbital nerve Extraconal	2.0 x 5.0	Superior orbitotomy	Uneventful
14	1961	Sharma[12]	M	15	Left proptosis Papilloedema	2 years	Superior maxillary nerve Intraconal	*	Biopsy excision	No light perception Intraoperative optic nerve damage
15	1968	Mortada[22]	F	35	Right proptosis Optic atrophy	1 year	Extraconal	3.0 x 3.0	Excision	No recurrence after 5 years
16	1968	Mortada[22]	F	60	Left proptosis Optic atrophy	2 years	Extraconal	2.0 x 3.0	Excision	No recurrence after 5 years
17	1968	Mortada[22]	F	50	Left proptosis Optic atrophy	7 months	Extraconal	5.0 x 3.0 x 1.0	Excision	Malignant neurilemmoma
18	1968	Mortada[22]	M	55	Right proptosis Visual loss	4 months	Extraconal	4.0 x 3.0 x 2.0	Excision	Malignant neurilemmoma Recurrence and lung metastasis after 9 months
19	1968	Mortada[22]	M	22	Right proptosis	15 years	Extraconal	*	Excision	Neurilemmoma of right ethmoid and maxillary bones
20	1968	Mortada[22]	F	19	Right proptosis Limited motility	5 years	Extraconal	*	Excision	Neurilemmoma of right ethmoid and maxillary bones
21	1968	Nakamura[23]	F	36	Left proptosis Blurred vision	1 year	Posterior ciliary nerve Intraconal	Walnut-sized	Excision	No proptosis Vision improved
22	1968	Nakamura[23]	F	20	Left proptosis	1 year	Posterior ciliary nerve Intraconal	*	Enucleation	Extensive tumor
23	1970	Mohan and Sen[10]	F	14	Right retrobulbar neuritis	10 days	Extraconal	*	Lateral orbitotomy	Proptosis and optic atrophy developed 3 years later
24	1971	Schatz[5]	F	27	Right orbital mass Proptosis	6 weeks	Supraorbital nerve Extraconal	4.0 x 5.0	Subfrontal approach	Sarcomatous transformation with metastasis
25	1973	Danziger[24]	F	57	Left proptosis Diplopia	3 months	Extraconal	3.0 x 2.0	Excision	Vascular mass appearance in angiography
26	1976	Imachi and Okuzawa[25]	F	50	Right proptosis	8 years	*	5.0 x 3.5 x 4.0	Excision	No recurrence after 6 months
27	1976	Oshima *et al*[26]	F	31	Left proptosis Visual loss	1 year	Intraconal	2.5 x 2.5	Excision	No proptosis
28	1977	Sinha *et al*[27]	F	35	Right proptosis	3 years	*	5.0 x 3.8	Partial exenteration	*
29	1977	Allman[28]	M	28	Left proptosis Blurred vision Diplopia	1.5 years	Extraconal	2.7 x1.9 x 3.3	Lateral orbitotomy	Visual acuity improved
30	1977	Allman[28]	F	67	Left proptosis Diplopia	10 months	*	1.5-in diameter	Lateral orbitotomy	No recurrence after 4 years
31	1977	Allman[28]	M	43	Left proptosis Squint	2 years	Extraconal	*	Transfrontal craniotomy	Total lateral rectus palsy
32	1977	Allman[28]	M	61	Left proptosis Blurred vision	3 years	Intraconal	*	Lateral orbitotomy	Vision improved
33	1979	Sen *et al*[29]	F	13	Left panopthalmitis Proptosis Pain	2 months	Ciliary nerve Intraconal	4.0 x 2.5 x 2.2	Enucleated	No recurrence after 2 years
34	1979	Kachole[30]	M	28	Left proptosis Diplopia Blurring	1 month	Intraconal	*	Excision	Malignant schwannoma
35	1980	Schmitt and Spoerri[31]	M	72	Right proptosis Diplopia	4 years	Extraconal	1.0 x 1.2 x 2.0	Transfrontal craniotomy	Total recovery
36	1982	Rootman *et al*[3]	M	60	Right proptosis Diplopia	36 years	Intraconal	*	Lateral orbitotomy	Full ocular function No recurrence
37	1982	Rootman *et al*[3]	F	54	Right central scotoma	1.5 years	Superior branch of oculomotor nerve; Intraconal	1.0 x 1.0	Piecemeal removal	Temporary partial ptosis and superior rectus paresis
38	1982	Rootman *et al*[3]	F	32	Diplopia in upgaze Right upper eyelid mass	8 years	Supraorbital nerve Extraconal	Occupy entire roof of orbit	Piecemeal removal	No recurrence
39	1982	Rootman *et al*[3]	M	24	Left orbital dull ache Left proptosis	*	Extraconal	*	Ethmoidectomy	Preoperative diagnosis: mucocoele
40	1982	Rootman *et al*[3]	M	31	Left lower orbital rim and eyelid mass Pain	7 months	Infraorbital nerve Extraconal	2.3 x 2.5	Excision	Complete recovery
41	1982	Chisholm and Polyzoidis[32]	F	34	Left proptosis Diplopia	1 year	Intraconal	*	Anterior craniotomy	Recurred 22 years later
42	1986	Zhang[33]	F	37	Left proptosis Blurred vision	3 years	Intraconal	3.0 x 2.0 x 1.5	Excision	Optic atrophy
43	1982	Horie *et al*[34]	F	41	Right proptosis Limited upgaze	3 months	Supraorbital nerve Extraconal	2.0 diameter	Subfrontal approach	Proptosis diminished Full ocular motility
44	1984	Konrad and Thiel[3]	M	84	Left proptosis Diplopia Headache	Many years	Extraconal	3.6 x 2.0 x 1.5	Excision	*
45	1986	Shields *et al*[35]	M	33	Headache Right proptosis	1 year	Intraconal	4.0 x 2.0 x 1.5	Lateral orbitotomy	No recurrence
46	1986	Cantore *et al*[36]	M	65	Proptosis	2 years	Extraconal	*	Excision	*
47	1986	Cantore *et al*[36]	F	32	Proptosis Limited ocular motility	9 years	Ciliary nerve Intraconal	*	Piecemeal removal	Decreased vision
48	1986	Jaais and Sivanesan[37]	M	35	Right proptosis Diplopia Blurring Pain	1 month	Extraconal	2.0 x 3.0	Biopsy	Metastatic malignant schwannoma
49	1988	Bergin and Parmley[38]	F	23	Right proptosis	4 months	Extraconal	3.0 x 2.5	Excision	*
50	1988	Bickler-Bluth[39]	F	38	Left proptosis Intermittent diplopia	Several months	Supraorbital nerve Extraconal	3.2 x 1.8 x 1.8	Anterior orbitotomy	No recurrence after 1 year
51	1988	Byrne *et al*[40]	M	41	Right proptosis Diplopia	2 years	Intraconal	3.8 x 2.0 x 1.5	Excision	*
52	1988	Byrne *et al*[40]	F	66	Left periorbital pain Blurred vision	5 months	Intraconal	2.2 x 1.8 x 1.2	Excision	*
53	1989	Sharma *et al*[41]	M	40	Right proptosis Headache	3 months	Intraconal	2.2 x 1.8 x 1.2	Superior orbitotomy	Meningioma was removed 3 months ago in the same eye
54	1990	Capps[14]	F	8	Right proptosis	3 years	Medial rectus Extraconal	Large	Biopsy	Mild proptosis Full motility
55	1990	Jensen and Bretlau[42]	M	22	Left proptosis	3 months	Infraorbital nerve Extraconal	3.0 x 1.0	Lateral orbitotomy	Recurrence after 3 years
56	1991	Asrani *et al*[43]	F	19	Right proptosis Blurred vision	2 years	Extraconal	6.0 x 4.0 x 4.0	Excision	*
57	1992	Eviator *et al*[9]	M	1	Right proptosis	3 weeks	Intraconal	2.8 x 2.5 x 1.2	Lateral orbitotomy	Malignant tumor No recurrence after 9 years
58	1993	Grover *et al*[4]	F	45	Right proptosis Diplopia Blurring	2.5 years	Intraconal	*	Lateral orbitotomy	No diplopia No proptosis
59	1994	Kanemoto and Okamoto[44]	F	70	Left proptosis Blurred vision	3 years	Short ciliary nerve Intraconal	*	Excision	Vision improved Pupil reaction loss
60	1997	Lam *et al*[13]	M	54	Right proptosis Diplopia	5 years	Supratrochlear Nerve Extraconal	3.4 x 2.5 x 2.2	Subbrow incision	Vision recovered No diplopia

of the tumor, the anterior, medial, cranial, or even panoramic approach has been adopted by some authors. Most patients have improvement in vision and ocular motility, with no recurrence, after total excision.

In summary, the variable location and presentation of orbital schwannoma can pose major diagnostic difficulties and challenges to clinicians. Orbital schwannoma usually grows slowly and produces a well-tolerated proptosis; however, significant rapid visual deterioration can occur if it is located in the intraconal space, especially when it is neglected. Although rare, orbital schwannoma has to be included as a preoperative differential diagnosis for both intraconal and extraconal orbital masses.

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