Orbital Hemangiopericytoma

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ABSTRACT

This report details a case of hemangiopericytoma in an 18-year-old male patient who was referred due to an orbital mass in the right eye. The patient presented with symptoms including blurred vision, proptosis (eye bulging), a pale optic disc, and a relative afferent pupillary defect (RAPD). Orbital computed tomography (CT) ordered by a referring ophthalmologist revealed an intraconal mass in the right orbit. The patient underwent an excision biopsy of the orbital mass through a lateral orbitotomy with osteotomy under general anesthesia. Significant hemorrhage was noted intraoperatively.

The mass was excised en bloc and sent for histopathological examination, which revealed a solitary fibrous tumor. Immunohistochemistry revealed hemangiopericytoma with malignant potential. The patient was referred to oncology, then radiotherapy for adjuvant external beam radiotherapy using intensity-modulated radiotherapy (IMRT).

Hemangiopericytoma or Solitary fibrous tumor is a rare mesenchymal tumor. Solitary fibrous tumors are uncommon, accounting for only 1.5% of tumors, and present as unilateral, painless masses that can cause proptosis.

Imaging is crucial for diagnosis and planning. Hemangiopericytoma appears as well-circumscribed lesions on CT and MRI. Histologically, it seems to have a staghorn-branched vascular pattern and stains positively on specific markers like CD34 and STAT 6.

Enbloc excision is the preferred treatment, often preceded by embolization to minimize bleeding. For high-risk cases, adjuvant therapy like radiotherapy may be utilized. Visual impairment affects the quality of life and mental health, noting that conditions like strabismus can lead to increased anxiety and depression. Strabismus surgery can enhance interactions and emotional well-being.

Orbital tumors vary by gender and age. The optimal approach to rare orbital tumors like Hemangiopericytoma is best determined by: detailed history, meticulous, cautious, comprehensive physical examination, and appropriate and thorough radiological evaluation. The justification for surgical intervention must consider the presence of a functional deficit, the risk of further progression, the suspected risk of malignancy, the lesion's location, and aesthetic considerations. Early detection, proper diagnosis, and management are crucial in preventing further complications.

Key words: Orbital hemangiopericytoma

The incidence of primary orbital tumors is very rare, with about 1 in 100,000 individuals occurrence. Most of these tumors are benign, though after the age of 60, there is an increasing risk for malignancy. In adults, primary benign orbital tumors are generally equally distributed. The upper outer quadrant is the most common site for these tumors, while those in the lower inner quadrant have a higher likelihood of being malignant.

The optimal approach to an orbital disease is best determined by a detailed history, and extensive review of symptoms, combined with proper physical examination and appropriate radiological evaluation

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to arrive at an accurate diagnosis and institute appropriate management.

Hemangiopericytoma, also known as a solitary fibrous tumor, is a rare mesenchymal tumor characterized by a genomic inversion at the 12q13 locus, leading to the fusion of the NGFI-A binding protein 2 (NAB2) and signal transducer and activator of transcription 6 (STAT6) genes. Orbital solitary fibrous tumors typically present as painless, slow-growing masses and generally have a favorable prognosis following surgical excision.

This paper presents a case involving an 18-year-old patient diagnosed with hemangiopericytoma. The case report aimed to increase clinical awareness and inform future diagnostic and management strategies for this rare tumor.

THE CASE

This is a case of an 18-year-old Filipino male, residing in Isabela who came in due to blurring of vision in the right eye.

The history of the present illness began 7 months prior to the consultation when the patient first noticed a progressive blurring of vision in the right eye. The patient initially consulted an optometrist, who prescribed glasses that did not alleviate the visual blurring. No medications were taken. Three months before the consultation, the blurring of vision persisted, prompting a visit to an ophthalmologist. An orbital CT scan was performed, revealing a lobulated soft tissue mass in the intraconal retrobulbar region, measuring approximately 2.4cm x 2.5cm x 2.1cm. The mass appeared closely associated with and indistinguishable from the slightly prominent right superior ophthalmic vein, showing similar enhancement in contrast studies. Additionally, the right globe was proptosed, the right optic nerve was compressed inferiorly, and the right superior rectus muscle was slightly compressed superiorly. Considerations include orbital venous varix, cavernous venous malformation, carotico-cavernous fistula and orbital lymphoma. The patient was referred to orbit and oculoplasty for further evaluation and management.

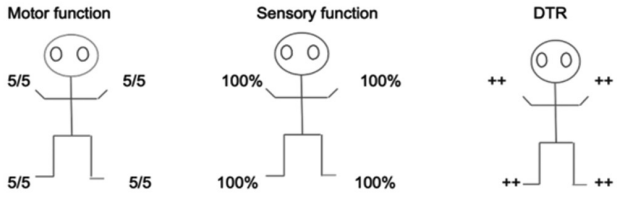
The patient is non-hypertensive, non-diabetic, and non-asthmatic, with no known comorbidities. Family history is unremarkable.

The patient is a non-smoker, nonalcoholic beverage drinker, with no history of prohibited drug use or unprotected sexual encounters. The patient was high school student at Nueva Ecija at the time of consult.

He denied any history of trauma, use of eyeglasses, and any history of ocular surgeries.

The patient was conscious, coherent, and not in cardiorespiratory distress. Vital signs were stable, with a blood pressure of 110/70 mmHg, a pulse rate of 79 bpm, a respiratory rate of 17 breaths per minute, a temperature of 36.5°C, and a weight of 54 kg.

CN I: no anosmia
CN II (+) Relative afferent Pupillary Defect, right, Pupil Reactive to Light, left
CN III: no limitation in all gazes
CN IV: no limitation in intorsion
CN VI: no limitation in abduction
CN V: symmetric facial sensation
CN VII: no facial asymmetry or weakness
CN VIII: intact hearing
CN IX, X: uvula midline, (+) gag reflex
CN XI: can shrug shoulders against resistance
CN XII: tongue midline on the protrusion



No signs of meningeal irritation. The rest of the neurologic exam was unremarkable.

Pre-operative ocular examination revealed a best-corrected visual acuity of only 15/400 on the right eye and 20/20 on the left eye. Other significant findings on the right eye included a 3 mm proptosis and positive for a relative afferent pupillary defect and a pale optic disc. The eye examination was unremarkable for the left eye. The slit lamp exam on both eyes was unremarkable.

CT scan of the orbit (A) Coronal and (B) Axial showing 2.4cm x 2.5cm x 2.1cm soft tissue mass that is located intraconally and retrobulbar which appears intimately related to and cannot be well delineated from the slightly prominent Right superior ophthalmic vein and exhibits similar enhancement in contrast study. There is proptosis of the right eye. The right optic nerve is compressed inferiorly and the right superior rectus muscle is slightly compressed superiorly.

Table 1. Ocular examination results

	Right eye	Left eye
Visual Acuity	**15/400** NIPH	20/20
External Eye	No lid swelling, No matting of lashes, non hyperemic conjunctiva, Pupils 2-3mm RTL	
Hertel's: 116mm	**18mm**	**15mm**
MRD 1	**3mm**	**4mm**
MRD 2	**5mm**	**5mm**
Slit lamp exam		
	Clear Cornea, Deep Anterior Chambers, No cells and flares, No lens opacity	
Fundoscopy	(+) ROR, Clear media, AVR 2:3, **Pale disc**, (-) Hemorrhages, (-) Exudates	Left eye- unremarkable

Fundus Examination

2A **2B**

Figure 2A(Left) Fundus findings of the right eye show Disc pallor with no hemorrhages and exudates. In comparison to **Figure 2B(Right)** Fundus finding of the left eye shows distinct disc borders with a CDR of 0.3 with no hemorrhages and exudates.

Radiologic Examination

Preoperative orbital CT scan

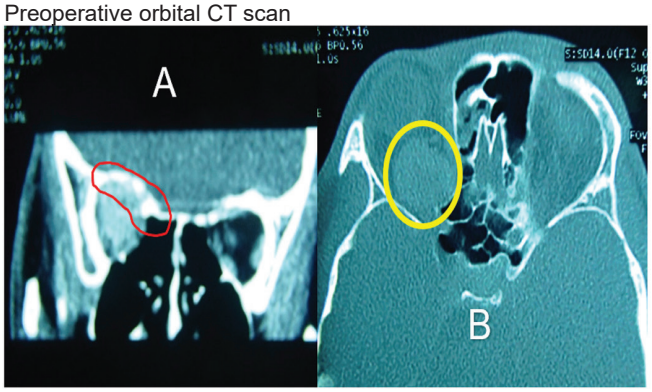


Figure 3A & 3B. CT scan of the orbits
Figure 3A shows some remodeling of the superior orbital wall
Figure 3B Axial view of the orbit showing a well-delineated retro bulbar mass measuring 2.4cm x 2.5cm x 2.1cm that appears to be closely related to the Right superior ophthalmic vein

There was consideration for further radiologic evaluation, in particular, magnetic resonance imaging (MRI) of the orbits. However, this was not done to focus financial resources on the surgery.

Preoperative Impression and Contemplated Surgical Procedure

According to the study of Khan, et al., the risk of blindness after orbital surgery is reported at around 0.84% and 24%, and it is more common with the orbital apex, with the highest incidence associated with vascular tumors, which was found in the present case.

Hence, the patient and his family were advised regarding the risks and possible complications of surgery, especially visual loss. The patient presented with preoperative poor vision ('blind' eye by legal definition) and signs of compressive optic neuropathy due to the orbital mass.

Literature states orbital surgery complication rates at 12.4%, with the highest rates for lateral orbitotomy at 35%; these can range from injury to extraocular muscles, globe rupture, orbital hematoma, and loss of vision.

Pre-operative planning was done to determine the surgical approach with the least possible complications.

The patient underwent excision biopsy of orbital mass through Lateral Orbitotomy with Beveled Osteotomies, Right Orbit, under General Anesthesia

After anesthesia induction and local periorbital subcutaneous injection, a corneal protector was applied to the globe. A 1.5-cm lateral canthal incision

beginning 10mm from the lateral canthal was made and dissection proceeded to the lateral orbital rim. The periosteum was incised and the lateral orbital wall was exposed. The temporalis muscle was retracted as necessary. The inferior and superior extent of the osteotomies were marked. Beveled osteotomies were made using a customized rotating bone saw. One (1) mm holes (Figure 4A, light arrows) were drilled superior and inferior to each planned osteotomy for refixation should further stabilization be desired. The lateral orbital bone was removed (Figure 4B). The orbital mass was localized (Figure 4C, white arrow) and excised en bloc (Figure 4D). Significant hemorrhage i.e. >40 pieces of soaked 8 x 10 gauzes amounting to blood loss of 1.1L (Figure 4E)The lateral orbital rim was repositioned and stabilized (Figure F). Layer-by-layer wound closure (Figures 4G & 4H). Placement of Frost suture (Figure 4I)

Differential Diagnosis

Depending on the mass's location—such as retrobulbar or intraconal—all of the following conditions can present with symptoms like blurring of vision, proptosis, and, if the optic nerve is involved, optic disc pallor.

Rhabdomyosarcoma may cause blurring of vision, proptosis, and optic disc pallor. It is typically located in the intraconal or retrobulbar regions. These tumors are usually well-circumscribed and represent the most common primary malignancy of the orbit.

Schwannoma can also present with blurring of vision, proptosis and optic disc pallor. It is often found in the extraconal or intraconal areas and is generally well-circumscribed, though it accounts for only about 1% of orbital tumors.

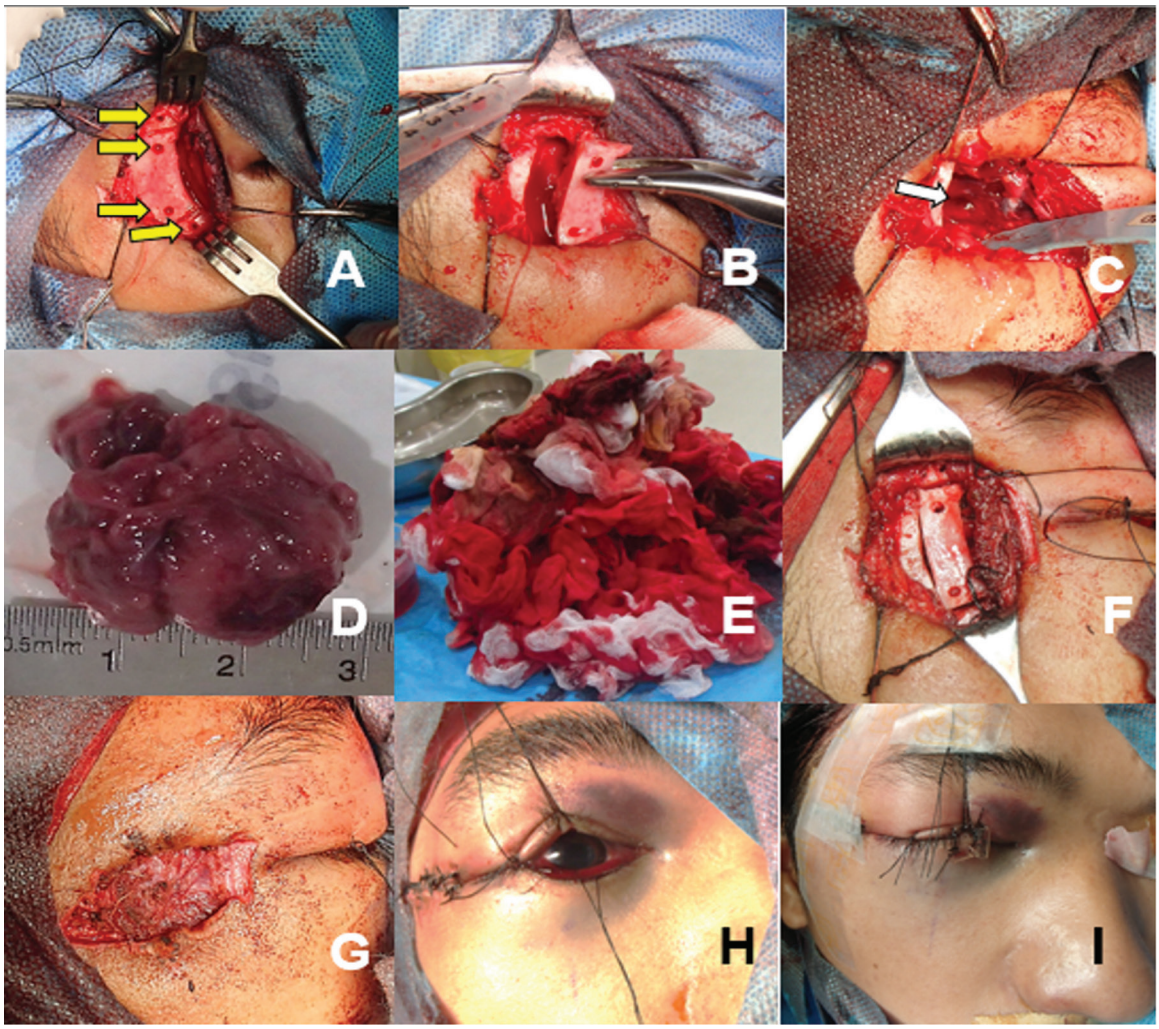


Figure 4. The image above shows the intraoperative findings of the excision biopsy of intraorbital mass through lateral orbitotomy with beveled osteotomies, Right Orbit, under General Anesthesia

Isolated Neurofibroma may present with similar symptoms—blurring of vision, proptosis, and optic disc pallor. These tumors are primarily located in the intraconal region but can also be found in the extraconal area. They are typically well-circumscribed but represent only 0.6-2.4% of orbital tumors.

Solitary Fibrous Tumor also presents with blurring of vision, proptosis, and optic disc pallor. It is usually located in the retrobulbar region and is generally well-circumscribed, though it is very rare among orbital tumors.

Cavernous Hemangioma may present with blurring of vision, proptosis, and optic disc pallor. These tumors are generally found in both the extraconal and intraconal regions, are well-circumscribed, and are the most common primary orbital tumor.

Pre-operative Impression: Orbital Mass T/C Cavernous Hemangioma, Right

Histopathologic Exam

A tan-gray, rubbery piece 2cm x2cm x 2cm upon reconstruction. The external surface is gray with focal areas of adhesion. There is central dark brown discoloration on the cut surface measuring .7cm x .8cm. Serial sections show tan-gray to brown surfaces.

Microscopically

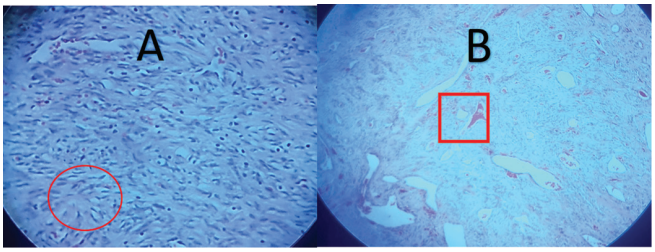


Figure 5 A shows the haphazardly arranged spindle to oval cells to round nuclei. **B** shows the scattered dilated, staghorn horn-like vessels

A tumor composed of the haphazardly arranged spindle to oval cells with oval to round nuclei disposed on a fibro collagenous stroma. Some oval cells exhibit clear cytoplasm. Scattered dilated and staghorn-like vessels are seen.

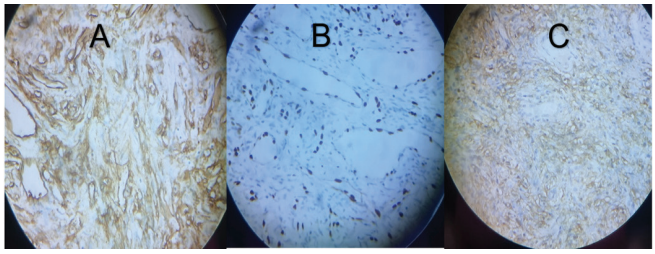


Figure 6 A, **B**, and **C** show Diffusely positive staining to CD34, STAT6, and CD99 respectively.

Table 1. Summary of differential diagnosis

	Case	Rhabdomyosarcoma	Schwannoma	Isolated Neurofibroma	Solitary Fibrous Tumor	Cavernous Hemangioma
Age	18yo	Pediatric- 5-7year old	Young to Middle age	3rd to 5th decade of life	Middle age	Middle age
Location Of Mass	Intraconal	Extraconal/ **Intraconal**	Intraconal<Extraconal	Retrobulbar	Extraconal/ **Intraconal**	**Intraconal**
Mass appearance on Imaging	Well circumscribed	Well circumscribed	Well circumscribed	Well circumscribed	Well circumscribed	Well circumscribed
Incidence		Most common primary orbital malignancy	1% of orbital tumors	0.6-2-4% of tumors	Uncommon	**Most common primary orbital tumor**
VA	Decreased	+	+	+	+	+
Proptosis	(+)	+	+	+	+	+
Optic Disc Pallor	(+)	+	+	+	+	+

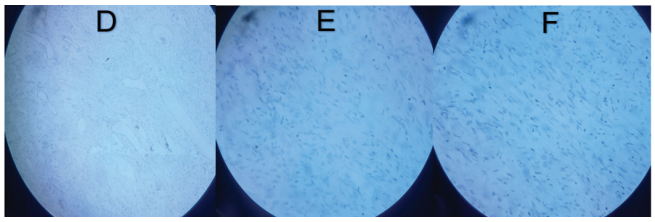


Figure 6 D, E, and **F** show CD31, S100, and Desmin respectively which are Negative to the cells of interest.

Histopathology and Immunohistochemistry revealed hemangiopericytoma with malignant potential.

The histopathology and immunohistochemistry results prompted a referral to Oncology. Oncology emphasized the limited data on adjuvant chemotherapy for hemangiopericytoma and suggested postoperative magnetic resonance imaging and referral to radiotherapy.

Post operative Radiologic Examination

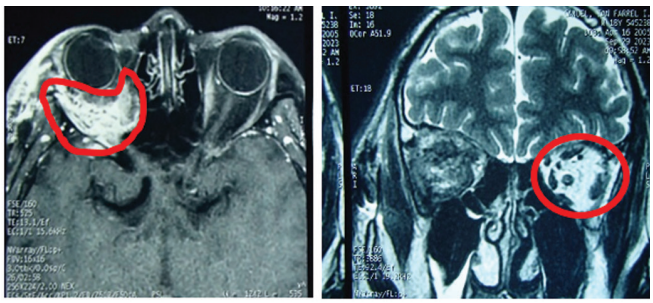


Figure A Figure B

Figure 7 A & **B** Outline the iso to hyperintense focus that almost occupies the intraconal and extraconal spaces of the orbit.

Post-operative Contrast-Enhanced Orbital MRI Done at the Institution

MRI of the orbit (A) Axial and (B) Coronal shows T1 isointense, T2 iso-to hyperintense focus with multiple intralesional signal voids almost completely occupying both the intraconal and extraconal spaces of the right orbit, particularly the apex and lateral aspect of the mid to anterior segment. There is medial displacement of the ipsilateral optic nerve while the ipsilateral lateral rectus muscle is not delineated. The lacrimal gland is displaced superiorly. There is a surgical bone defect in the right zygomatic process.

This was signed out as, CONSIDER TUMOR RECURRENCE OF KNOWN PRIMARY, Right orbit

Radiotherapy as Adjuvant Treatment

Radiation oncology recommended radiotherapy. However, the patient opted to undergo the procedure at their provincial hospital. He underwent external beam radiotherapy utilizing intensity-modulated radiotherapy (IMRT), with a dose prescription of 50 Gy to the right orbit. The treatment regimen was fractionated into 2.0 Gy per fraction and administered over 25 sessions from October 23 to December 5, 2023. The patient completed the prescribed course of radiotherapy and tolerated the treatment well.

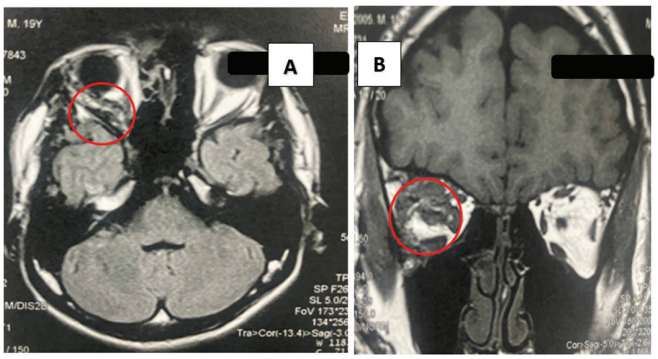


Figure 8 A & **B** show an MRI of the orbit with Coronal and Axial views demonstrating an irregularly shaped retrobulbar abnormality that projects toward the region of the right apex

Post-Radiotherapy Orbital MRI Done at Another Institution

Orbital MRI revealed a 1.7 cm heterogeneously enhancing mass in the retrobulbar region. This mass predominantly consists of fibrotic tissues, which are likely due to post-operative granulation changes.

Postoperative Course in the Wards

Immediate postoperative visual acuity on the right eye was No light perception, Extraocular movements were -4 on abduction and -3 on adduction with pupils 7-8mm Nonreactive to light. There is periorbital edema but no signs of infection. Since compressive optic neuropathy due to hemorrhage, inadvertent damage to the optic nerve cannot be totally ruled out, the patient was started on Methylprednisolone 250mg IV every 6 hours for 3 days.

Post Operative Ocular Examination

The visual acuity in the right eye failed to improve and remained No Light Perception, while the visual

acuity in the left eye remains 20/20. Post-operatively, there is a +4 esotropia in the right eye. Forced duction testing of the right eye reveals tightness in the medial rectus muscle. According to the study by Khan, et al, the risk of blindness after orbital surgery is around 0.84% to 24% and it is more common with orbital apex tumors. The reason could either be direct nerve injury or ischemic damage to the optic nerve or retina or central retinal artery occlusion secondary to excessive manipulation and bleeding in lateral orbitotomy.

DISCUSSION

This case report focuses on orbital hemangiopericytoma, a rare mesenchymal tumor also known as a solitary fibrous tumor. Tumors in the orbit can vary significantly by gender and age. Among infants and children, the most common primary malignant tumors are rhabdomyosarcoma and retinoblastoma. The former is more frequent in males.

In adults, hemangiomas are the most common benign primary orbital tumor and have been equally distributed among the sexes. Meningiomas are the second most common benign tumor affecting the orbit, which is common among the female gender. Dermoid cyst, which is also a benign tumor, is also often seen and affects both sexes equally. Orbital lymphomas are the most common primary malignant tumor and have a higher prevalence among females. Among the elderly, metastasis from the breast, lung and prostate is more prominent. Primary orbital tumors, which include schwannomas, may differ in sex distribution depending on tumor type.

Orbital involvement was first described in 1995. Hemangiopericytomas originate from the fibroblasts of mesenchymal cells and are notably infrequent, with an incidence of approximately 3.77 cases per 10,000,000 individuals, according to Seo, et al.[2] Solitary fibrous tumors of the head and neck are particularly rare, representing only about 6% of such tumors, and orbital solitary fibrous tumors account for a mere 1.5% of cases at all sites.[3] According to the study by Thompson, et al.[3] only 2 million patients per year are diagnosed with SFTs.

Orbital solitary fibrous tumors a to have no gender predilection.[4] It is more common that solitary fibrous tumors will present among younger age groups than in the 5th—7th decades.[4]

Solitary fibrous tumors are slow-growing tumors that may affect any age group. They usually present as painless unilateral proptosis and present for 6-12 months before diagnosis.[4,5] Blurring of vision, ophthalmoplegia, ptosis and diplopia are rare clinical conditions.[4] As the mass progresses, there is an increased mass effect and possible exposure to keratopathy and even optic atrophy in later findings.

Radiography is important in localizing, determining the size, planning surgical interventions and postoperative monitoring. Orbital solitary fibrous tumor appears as a well-circumscribed, solitary ovoid mass on both CT and MRI and appears as contrast enhancement due to its high B vascularity On a CT scan, SFTs appear as a well-circumscribed ovoid lesion, which can either be isodense or hyperdense compared to the cerebral cortex. On MRI, it appears as B homogenous isointense or hypointense signal intensity on T1 and mixed intensity on T2.

The majority of solitary fibrous tumors are associated with a genomic inversion at the 12q13 locus, leading to the fusion of the NGFI-A binding protein 2 (NAB2) and signal transducer and activator of transcription 6 (STAT6) genes.[6] Orbital solitary fibrous tumors typically present as unilateral, well-circumscribed masses that exhibit contrast enhancement on CT and MRI due to their high vascularity.[7,8]

Microscopically, solitary fibrous tumors are identified as a staghorn-branched pattern of thin-walled sinusoidal-like vasculature. Solitary fibrous tumors stain positive for CD34 and can show high sensitivity to nuclear STAT6 immunohistochemistry staining. Additional histochemical stains may include vimentin, BCL2, and CD99. In contrast, solitary fibrous tumors are usually not stained for S100 protein, cytokeratins, smooth muscle actin or epithelial membrane antigen.[6,9,10]

When intervention is necessary, surgical excision en bloc is the preferred treatment approach for both primary and recurrent cases. This method helps reduce the risk of recurrence, local aggressive behavior, or malignant transformation that may occur years later. Pre-operative intravascular embolization is recommended to minimize intraoperative bleeding.

For instance, Demura, et al. used a coil and n-butyl-2-cyanoacrylate (NBCA). They embolized feeder vessels arising from the ophthalmic artery with coil and the infraorbital artery with NBCA. Preoperative endovascular embolization allowed for complete tumor removal without significant intraoperative hemorrhage. The surgical approach depends on size, location, and tissue infiltration.[11]

For high-risk cases, adjuvant therapies such as radiotherapy may be considered. Stereotactic radiosurgery, which delivers precise doses of radiation based on a carefully-designed treatment plan can be particularly beneficial as an adjuvant treatment following incomplete resection or for recurrent orbital solitary fibrous tumors.[12,13]

In the study of Konstantin, et al.,[14] a novel technique called pencil beam proton therapy was used as adjuvant treatment for a patient who underwent surgical removal of hemangiopericytoma and had already undergone conventional radiotherapy. In their study, the patient underwent pencil bean PT as the fourth course of irradiation. After 9 months of therapy, it showed a complete tumor response with signs of brain toxicity. This showed the possibility of a new therapy with a promising result and manageable toxicity.

Vision impairment can lower the quality of life. Quality of life encompasses different aspects, including emotional health, social interactions, vision-related concerns and convenience. Research has demonstrated that ocular trauma has far deeper consequences for the person than just vision loss. A person's total impairment from blindness in one eye is 24%; if both eyes are blinded, the impairment is 85%.[15] In addition to being a chronic condition in and of itself, vision loss exacerbates the effects of other chronic conditions. Individuals who suffer from vision impairments along with other illnesses or conditions are more likely to experience difficulties completing tasks and to report feeling unwell. Furthermore, other conditions, such as vision rehabilitation, which aims to enhance the functionality and quality of life for people with vision impairments, may have an impact on the management of eye disease.[16]

Individuals with strabismus are ten times more likely than those without the condition to experience clinical depression or anxiety. Strabismus also increases the risk of mental health problems. Individuals who have strabismus are more likely to experience increased social anxiety and avoidance, worry about how they look, and have a pessimistic attitude toward life. In addition, they might experience low self-esteem, shy away from particular situations, and even consider quitting school. Nonetheless, by enhancing social interactions and emotional well-being, strabismus surgery can improve patients' overall well-being. Following surgery, they frequently report improved eye contact and more pleasant social interactions. Hence, the patient was referred to the pediatric and strabismus surgeon, and currently, it is planned for vertical muscle transposition and medial rectus recession.

CONCLUSION

Tumors in the orbit can vary significantly by gender and age. Hemangiomas are the most common benign primary orbital tumor, followed by meningiomas. Depending on the mass's location—such as retrobulbar or intraconal—all of the following conditions can present with symptoms like blurring of vision, proptosis and, if the optic nerve is involved, optic disc pallor.

The optimal approach to an orbital disease is best determined by a detailed history, and extensive review of symptoms, combined with proper physical examination and appropriate radiological evaluation to arrive at an accurate diagnosis and institute appropriate management, especially in Hemangiopericytoma, which is a rare form of solitary tumor. Even though they are rare, they should taken into account due to their sluggish growth and the ability to have a major impact on vision.

Imaging methods and histological analysis are necessary for diagnosis. Removal of the entire tumor with preoperative embolization helps reduce bleeding and improves surgical outcomes. Adjuvant therapies and innovative methods like Pencil beam proton therapy should be taken into consideration for patients with a high risk of recurrence.

All things considered, Orbital Hemangiopericytomas are uncommon and require specific knowledge and care. Improvements in radiotherapy and surgery continue to enhance patient outcomes and provide hope for better management of this difficult tumor.

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