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# Masquerading Orbital Cavernous Venous Malformation in a Young Female, at Tertiary Hospital, North East, Nigeria- A Case Report

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## ABSTRACT

***Background:*** *Orbital Cavernous hemangiomas are the most common benign, slow growing primary orbital tumor in adults, frequently presenting in middle -age women with progressive painless proptosis. The tumour, represent 5% of orbital masses, sometimes they are described as solitary, encapsulated venous-lymphatic malformations. It typically arises within the muscle cone in 80% of the cases, causing visual impairment or diplopia due to pressure on the optic nerve. Restriction of ocular motility and orbital pain are also documented in several cases even though most cases appear asymptomatic due to its slow progressing characteristic.*

***Method:*** *Informed consent was obtained from the patient, and ethical approval obtained from the Hospital Ethical Committee. Patient was a 25year old female who presented with a history of painless and progressive right eye protrusion of 4/52 duration, this progresses to become painful with gradual loss of vision, redness and tearing associated with severe generalized headache. Ocular examination reveals unaided visual acuity of 6/6 in both eyes at presentation. However, VA in the RE progressively worsens to CF @3m and no improvement with best correction, lid ecchymosis and severe conjunctival chemosis with gross axial proptosis measuring 28mm, and restriction of ocular motility in all directions.*

*Dilated fundoscopy reveals pink disc with cup disc ratio of 0.3 in BE, elevated and blurring of disc margin with dilated blood vessels in the RE. Intraocular pressure was 18mmHg and 16mmHg in RE and LE respectively. No associated vascular malformations were identified following systemic review.*

*Patient had CT scan which showed a lobulated heterogenous hyperdense mass with multiple calcification and hypodense areas occupying the retrobulbar space. The mass is inseparable from the extraocular muscles and obscures the optic nerve.*

***Result:*** *She had modified exenteration and histology of the tissue which reveals characteristic features of capillary haemangioma.*

***Conclusion:*** *Computed tomography (CT) is one of the primary imaging modalities used to evaluate orbital tumors and vascular lesions, however, in this case definitive diagnosis was made only following histology.*

**Keywords**
Haemangioma, Exenteration, CT scan, Proptosis.

## Introduction

Orbital Cavernous hemangiomas (or cavernous venous malformations) are the most common benign, slow growing primary orbital tumor in adults, frequently presenting in middle -age women with progressive painless proptosis. The tumour represent 5% of orbital masses [1], sometimes they are described as solitary, encapsulated venous-lymphatic malformations [2]. It typically arises within the muscle cone (intraconal) in 80% of the cases [3], causing visual impairment or diplopia due to pressure on the optic nerve. Restriction of ocular motility and orbital pain are also documented in several cases even though most cases appear asymptomatic due to its slow-progressing characteristic [4].

Histologically, they are well-circumscribed encapsulated masses containing a complex network of dilated vascular channels lined by mature, flattened endothelial cells surrounded by abundant fibrous stroma [2].

Computed tomography (CT) and magnetic resonance imaging (MRI) are the primary imaging modalities used to evaluate orbital tumors and vascular lesions [5]. CT shows a well-defined, oval or rounded mass, usually located inside the muscle cone while MRI typically reveals progressive filling with contrast, where parts of lesion remain non-enhanced while others become brightly enhanced [5].

A and B scan are also helpful in making diagnosis especially where MRI and CT imaging are not readily available.

A-scan showed high reflectivity with regular structure, and moderate attenuation of echoes while B-scan reveals encapsulated, medium high reflectivity, and no signs of internal vascularization and no calcifications

Management of patients depends on the presentation, asymptomatic lesions do not require treatment and they may be observed, however surgery is indicated for symptomatic patients with optic nerve compression, marked disfigurement from proptosis and visual loss. Various types of surgical approaches have been proposed and exploited in different situations according to the anatomical location and nature of the lesion [3]. Anterior orbitotomy is increasingly applied for almost 60% of hemangioma removal located either at the extraconal or intraconal side. For lesions lying over the posterior third of the orbit, lateral and transcranial orbitotomy represent a more suitable approach than other methods despite a higher risk of developing surgical complications [3].

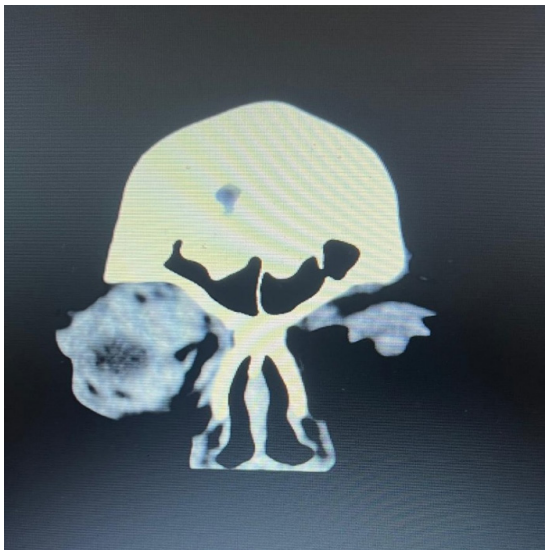
## Case Presentation

Patient was a 25year old female who presented with a history of painless and gradual progressive right eye protrusion of 4/52 duration, this progresses to become painful with gradual loss of vision, redness and tearing associated with severe generalized headache. There were no diplopia and gaze abnormalities and no other ocular symptoms, systemic history was also nil of note.

She had similar incidence at the age of 7years and was operated in a tertiary hospital but there was no documented histology report.

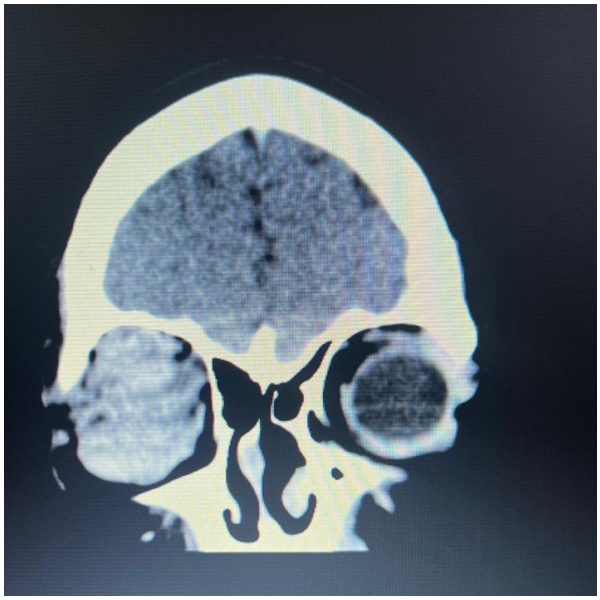
General physical examination was essentially normal. Ocular examination reveals unaided visual acuity of 6/6 in both eyes at presentation. However, VA in the RE progressively worsens to CF @3m and no improvement with best correction, lid ecchymosis and severe conjunctival chemosis with gross axial proptosis measuring 28mm using hertel exophthalmometer with restriction of ocular motility in all directions of gaze. Anterior segment structures and pupillary reflexes were essentially normal. Findings on the LE were also normal. Dilated funduscopy reveals pink disc with cup disc ratio of 0.3 in BE, elevated and blurring of disc margin with dilated blood vessels in the RE but no optic disc atrophy. Intraocular pressure was 18mmHg and 16mmHg in RE and LE respectively. No associated vascular malformations were identified following systemic review.

She had orbital CT where the serial axial pre and post contrast images with coronal and sagittal reformation showed a lobulated heterogenous hyperdense mass with multiple calcification and hypodense areas occupying the retrobulbar space. It demonstrated moderate enhancement on post images, the mass was inseparable from the extraocular muscles and obscures the optic nerve, resultant anterior and lateral protrusion of the globe, no bony erosion or destruction seen. An impression of right retro-orbital mass with differentials of Rhabdomyosarcoma, Lymphoma and Orbital dermoid was made. See radiograph 1 and 2. Blood investigation requested includes, Full blood count, grouping and cross matching, electrolyte, urea and creatinine, fasting blood sugar which were essentially normal.



**Radiograph 1:** Contrast enhanced coronal reformatted CT scan of the brain at the level of the orbits showing an enhancing mass lesion inseparable from the medial and superior recti muscles displacing the globe laterally.

Patient undergoes right eye modified exenteration and histology of the tissue.



**Radiograph 2:** Contrast enhanced coronal reformatted CT scan of the brain at the level of the orbits showing an enhancing mass lesion occupying almost the entire orbital cavity inseparable from the medial and superior recti muscles.

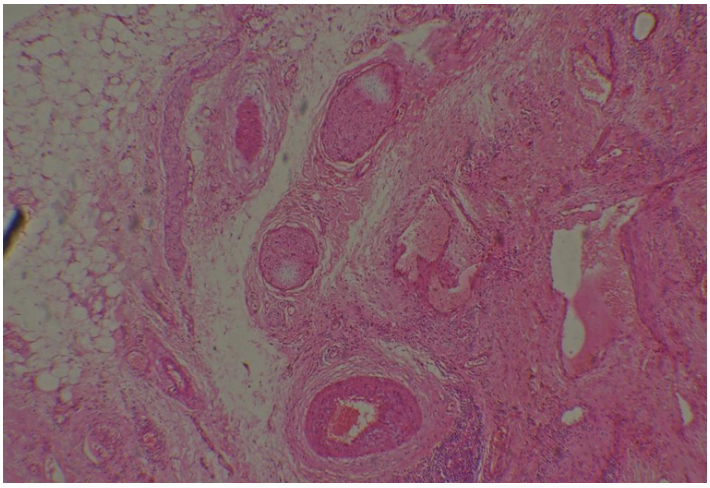
## Surgical Procedure

Patient cleaned and draped in supine position under general anaesthesia. An incision is marked on the eyelid approximately 2mm from the eye lash line. Tractional suture using 4-0 silk are placed through the tarsal plates, this helps to maneuver the contents of the orbit. An incision is made through the skin and orbicularis oculi muscles and dissected down to the bony orbital rim. The periosteum is elevated from the bony wall of the orbit $360^0$ starting from the bony orbital rim using the periosteal elevator. The soft tissues were dissected down to the apex, the optic nerve, extraocular muscles and tumour mass behind the optic nerve head were severed using curved scissors and elecrocautery to remove the contents completely. Homoestasis was achieved using cautery, the two lids were closed using chromic 6/0 sutures and a drained was inserted which was removed 72 hours after when no more fluid was draining. There was minimal blood loss during the procedure and so patient was not transfused with blood. She was placed on daily wound dressing, then alternate days until the wound site was cleaned and dry. She was also placed on systemic oral antibiotics for two weeks, analgesics for one week and povidone eye ointment. Patient was discharge from the hospital one week after surgery and she is currently doing well as at the time of this report. The tissues removed preserved in formalin bottle and sent to the laboratory immediately for histological diagnosis.

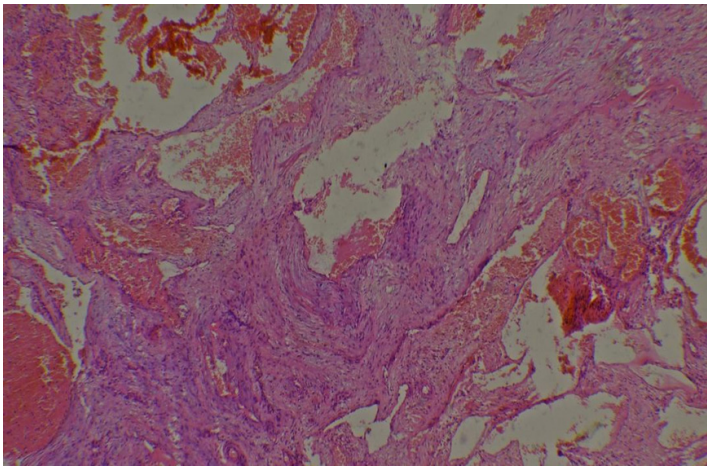
## Histology Report

Gross histology reveals an orbital specimen, measures 6.5x5x4 cm. opening reveals an eye encased by a greyish brown tissue having areas of haemorrhages and tiny congested spaces.

Microscopy sections showed a fibrous stroma, within which were numerous vascular channels, most of which were thick walled and congested. Surrounding these vascular spaces were dense lymphocytic aggregations with attempt at follicle formation. Areas of haemorrhage and unremarkable adipocytes were also noted. These vascular proliferations were seen surrounding the eye ball. Overlying unremarkable skin and skeletal muscle bundles were also noted, no evidence of malignancy, these features were consistent with haemangioma. See histology slides 1 and 2.



**Histology slide 1:** H&EX100: Photograph showing thick walled vascular channels, adjacent dilated and congested ones.



**Histology slide 2:** H&EX100: Photograph showing multiple vascular channels, some of which are dilated and congested CT SCAN.

## Discussion

Cavenous haemangiomas are slow growing benign tumours that present with gradual onset and slowly progressive proptosis which eventually leads to visual loss due to optic nerve compression especially in lesion located at the orbital apex. This study reports a case of 25year old female who was diagnosed with Cavenous haemangioma after modified exentheration. This correspond with

previous studies [3,4] that reports a higher predilection in female gender but older age group than our patient, ranging from 40-50 years, probably due to circulating estrogen/progesterone levels affecting the progression of the tumour [6].

In the past few years several different surgical procedures have emerged and evolved which includes anterior and lateral orbitotomy with transcutaneous, transconjunctival, or transcranial approaches, they have proved to be the favored methods based on the anatomical location, characteristics of the lesion, and the operating surgeon (ophthalmologist or neurosurgeon) [7,8].

Li-Wei Cheng et al. carried out a retrospective study on adjunctive aspiration technique for the surgical management of deep orbital cavernous hemangioma, where he reviewed11 patients who had sub-brow orbitotomy and inferior transconjunctival approach in two cases. All the tumors were removed successfully after needle aspiration of 1–3 cc of intralesional blood to reduce the tumor size. This approach shortened the operation time and reduced wound scar due to no additional bone structure removal and reconstruction, as well as lower risk of developing severe complications such as permanent visual loss or esthetic facial problems [3].

In contrast, our patient had modified exentheration because the CT scan result failed to give the exact definitive diagnosis of capillary haemangioma and patient condition was deteriorating. Even though, CT and MRI are the primary imaging modalities used to evaluate orbital tumors and vascular lesions, radiological differentiation from other solitary orbital masses can still be challenging at times [9]. Some benign and malignant orbital tumors (meningiomas, schwannomas, neurofibromas, malignant fibrous histiocytomas, solitary fibrous tumors, fibrosarcomas, and metastases) appear morphologically identical to orbital cavernous hemangiomas especially on non-contrast CT and/or MR imaging [9].

Once diagnosis is confirmed, patient can have surgical procedures like orbitotomy with adjunctive aspiration technique or endoscopic approaches which are less destructive and have better visual preservation.

Conclusion- Special emphasis should be given to contrast imaging techniques like CT and MRI as well as ocular A and B scan for making definitive diagnosis in orbital masses, this will enhance the choice of surgical procedure and approach, as orbitotomy give patient better chances of visual preservation and excellent cosmetic appearance than subtotal exentheration.

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