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Management of Complex Retrobulbar Orbital Tumours, Our Institutional Experience

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Abstract

The orbital tumours coming to neurosurgical team are often those, which are referred by ophthalmological units when they consider it to be complex in nature that is retrobulbar tumours complicated by extraorbital and intracranial extensions. *Methods*: clinical records of 23 patients operated for retrobulbar orbital pathologies in department of neurosurgery, Bangalore medical college and research institute from 2011 to June 2016 were reviewed and analysed. Patient's demographics, symptoms and signs at presentation, and histologic diagnosis were recorded. *Results*: Twenty three patients were operated, of which 12 were men and 11 women, and the age of patients ranged from 11 to 65 years and mean age was 43 years. Fourteen patients had primary intraorbital tumours, 8 patients had secondary orbital tumours extending from other adjacent contiguous bones or structures and 1 patient had metastatic tumour from ovarian neuroendocrine carcinoma. *Conclusions*: Orbit being a complex anatomical entity, management of complex tumours poses a challenging surgical problem. Although a significant percentage of these tumours are treated by the ophthalmologist alone, proficiency with a multitude of approaches and collaboration with a neurosurgeon is often required, especially for tumours that are located deep within the orbit, are large, or have an intracranial extension. Technical advances and modifications in surgical techniques along with involvement of plastic surgeons have decreased surgery-related morbidity and involvement of radiation oncologist to the team for adjuvant radiotherapy/chemotherapy would be beneficial for prolonged recurrence free and better quality of life.

Keywords: Orbital Tumours; Proptosis; Lateral Orbitotomies; Superior Orbitotomies; Craniofacial Excisions.

Introduction

The orbit is an anatomically complex structure housing the globe; extra ocular muscles; fat; and vascular, nerve, glandular, and connective tissues. This complex anatomical area was first described by Whitnall to resemble a pear, with its widest aperture anterior and narrowing posteriorly. The bony orbit embryologically develops from the mesenchymal cells surrounding the optic vesicle. Composed of 7 bones, a normal adult orbit holds a volume of 30 mL with a medial length of 45 mm, a width of 40 mm, and a height of 35 mm at its most anterior point.

Since the orbit is a relatively small anatomical area with little wasted space, space-occupying lesions that increase orbital volume may result in proptosis of the globe and may adversely affect visual and extraocular muscle function. However, Lin and colleagues reported that proptosis of less than 4 mm might go undetected, obscuring occult pathology [1].

A basic understanding of orbital anatomy is critical in evaluation of orbital neoplasms. Not only can site of involvement guide the differential diagnostic considerations, but accurate descriptions

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of lesion location, involved structures, and extent of dissemination can facilitate proper treatment planning. The orbit can be divided into the ocular compartment or globe, the muscle cone, and intraconal and extraconal spaces. Six extraocular muscles control ocular movements; all but the inferior oblique muscle constitute the muscle cone. The muscle cone converges at the orbital apex, forming a tendinous ring (annulus of Zinn). Through this ring enters the optic nerve, oculomotor nerve (superior and inferior divisions), abducens nerve, nasociliary branch of the ophthalmic nerve, and the ophthalmic artery. The muscle cone separates the intraconal and extraconal spaces.

Orbital tumours may be primary, meaning they originated from orbital structures, or secondary, meaning they extend into the orbit from adjacent structures and third group of tumours, involving the orbit are metastatic [2]. These tumours are variable in their biological nature and in their location [3,4]. Orbital tumours are also divided anatomically, into intraconal, extraconal, and intra-canalicular. This distinction is made on the basis of the tumour's relationship with the muscle cone, with the intra-canalicular group of tumours being those lesions that are at least partially within the optic canal [5]. Patients with primary or secondary orbital tumours usually present with exophthalmos, pain, diplopia, swelling, tearing, and blurred vision.

Imaging studies used in the diagnosis and management of orbital tumours have changed dramatically in the last few decades. Ultrasonographic examination of the orbit is still helpful in the evaluation of cystic lesions and also for vascular lesions. Magnetic resonance imaging gives high-resolution images of the normal components of the orbit and of nonosseous lesions in three dimensions. In cases of osseous lesions, CT is the modality of choice, either alone or in combination with an MR imaging. The drawback to using CT in examining the eye is that it uses ionizing radiation and may produce cataract.Angiography is still the imaging modality of choice for vascular lesions, such as ateriovenous malformations and low-flow ateriovenous fistulas. It is also useful in cases of the more vascular orbital tumours (such as meningiomas) for defining the extent of vascularity prior to surgery. Cerebral angiography should be undertaken in any patient with pulsatile exophthalmia.

Surgery

There are two major types of surgical approaches to orbital tumours: the transorbital approaches, which are usually performed by an ophthalmologist [6] and the extraorbital approaches, which are best performed by a team that includes a neurosurgeon or a head and neck surgeon as well as an ophthalmologist [7]. In most instances, anterior lesions are treated via transorbital approaches, whereas lesions of the posterior third are best managed via extraorbital approaches [7,8]. There may be some instances, however, where this rule, cannot be applied. In some cases, posterior lesions can be approached via extended or combined transorbital approaches, and lesions of the middle third of the orbit are easily accessible via extraorbital approaches.

In 1888, Krönlein first described the lateral orbitotomy approach. The Berke-Reese modification of this approach used an extended canthotomy. In place of the curved incision used by Krönlein, Stallard altered the approach taking it into the upper lateral brow area. Avoiding the lateral canthal region was at the core of the Wright modification of the Stallard approach. A hemicoronal (or bicoronal) approach, also referred to as the coronal approach, has been used by Kennerdal and others [9]. Goldberg et al have popularized the transconjunctival approach and the transcaruncular approach [10].

The transcranial approach in which the orbital rim is preserved was initially developed by Dandy [11] and has been subsequently modified. The transcranial approach with orbital osteotomy was initially described by Frazier in 1913 [12]. and has since been modified several times [13]. This approach is useful in cases of tumours involving the medial orbit and the anterior skull base. The subcranial approach can also be useful in midline lesions involving the orbit, the anterior skull base, and the paranasal sinuses.

Material and Methos

Twenty three patients with orbital tumours were operated in department of neurosurgery, Bangalore medical college and research institute from 2011 to June 2016, 12 were men and 11 women, the age of patients ranged from 11 to 65 years and mean age was 43 years. One patient had lacrimal adenoma (Figure 1), three patients had lacrimal adenoid cystic carcinomas (Figure 2), one patient had intraorbital abducent nerve schwanoma (Figure 3), one patient had NF-2 with intraorbital intraconal sensory nerve schwanoma (Figure 4), three patients had orbital apex meningiomas (Figure 13), two patients had post-

traumatic orbital ateriovenous malformations mainly involving upper eye lids following penetrating injury, one patient had lower eyelid solitary plasmacytoma, one patient had orbital lymphoma (Figure 5), one patient had plexiform neurofibrma with orbital extension with pthysis bulbi, one patient had sphenoid wing meningioma with intraorbital extension (Figure 6), one patient had recurrent maxillary adenocarcinoma with intraorbital extension, one patient had sinonasal carcinoma with intraorbital and intracranial extension (Figure 7), one patient had frontal mucocele with intraorbital and intracranial extension (Figure 8), one patient had frontoorbital solitary fibrous dysplasia, one patient had fronto orbital Ewing's sarcoma (Figure 9), one patient had fronto orbital angiosarcoma (Figure 10), one patient had ethmoido-orbito-cranial mucormycosis (Figure 11) and one patient had orbital metastasis secondary to ovarian neuroendocrine tumour (Figure 12). The clinical features (Table 1), headache was the commonest symptom which was bothering patients, and progressive proptosis though present, patients reached after diplopia and loss of vision compelled them to consult.

Results

Based on the clinical presentation and radiological findings surgical approaches were planned for individual cases (Table 2 and 3). Lateral orbitotomy was used in one patient (Figure 1) and complete excision of tumour involving the lacrimal gland was done. The orbit was reconstructed and patient did not have any deficits. The histopathological examination findings were suggestive of lacrimal gland adenoma.

Three patients, who had intraorbital superolateral quadrant lesions (Figure 2) extending upto orbital apex and into optic canal. All these patients had complete loss of vision and proptosis in the involved eye. Fronto-temporo-orbital craniotomy was done in 2 patients and fronto temporo-zygomatico-orbital approach was used in one along with enucleation of the involved eye in all three case. The histopathology was suggestive of adenoidcystic carcinoma of lacrimal gland in all three patients. All three patients are still alive and have not had recurrence on follow up of 3 years duration.

Three patients of optic nerve sheath tumour (Figure 13) were operatred by fronto-orbito-zygomatico- temporal craniotomy, only partial excision was possible in one patient and when complete excision was achieved in other two patients, both developed 3rd nerve palsy and complete loss of vision in involved eye. The histopathological examination was suggestive of meningioma of optic nerve.

One patient of sphenoid wing meningioma (Figure 6) with intraorbital and optic canal extension was approached by fronto-temporo-zygomatico-orbital craniotomy and Simpson's grade 1 excision was achieved and superior and inferior orbital wall were reconstructed with osteosynthetic bioreabsorbable mesh. Patient had complete functional preservation of eye and acceptable cosmetic result. The histopathological examination revealed meningioma.

One patient who had right intraorbital intraconal orbital apex lesion (Figure 4), was approached by fronto-temporo-zygomatico-orbitotomy and complete excision of sensory nerve sheath tumour was achieved. Histopathology was suggestive of schwanoma. Patient has complete functional preservation of the eye.

One patient with right intraorbital abducens nerve schwanoma (Figure 3)[14] and was approached by right frontotemporal craniotomy and superior orbitotomy with preservation of the orbital rim and total excision of the tumour was done and superior wall of the orbit was reconstructed with osteosynthetic bioreabsorbable mesh. Patient had complete functional preservation of eye and superior cosmetic result. Histopathological examination of the tumour revealed Antoni type 'A' schwanoma.

Weber-Fergusons facial degloving approach with maxillectomy with inferiormedial orbitotomy and complete excision was done in one patient of recurrent vercucous adenocarcinoma of maxillary sinus with intraorbital extension and this patient had complete functional preservation of involved eye.

One patient with left orbital plexiform neurofibrma with pthysis bulbi and was operated by superior transorbital approach through upper eye lid transverse incision extending from medial to lateral canthus. The eye lids were reconstructed by local flaps.

One patient with right frontoorbital bulge (Figure 9) and was approached by frontoorbital craniotomy and wide excision of superior orbital rim. Histopathology was suggestive of Ewing's sarcoma and patient was subjected for postoperative radiotherapy. Another patient with right frontal diffuse expansile bony swelling and was approached by frontoorbital craniotomy and wide excision. Histopathology was suggestive of monoostotic

fibrous dysplasia.

An immunocompromised patient secondary to HIV infection with CD4 count of 800, had left orbital proptosis (Figure 5) with pthysis bulbi and was approach by extended medial-orbitotomy and complete excision of lesion and enucleation. Histopathological examination was suggestive of non-hodgkin's lymphoma. Patient was referred to oncological unit for appropriate chemotherapy and this patient is lost to follow up.

One patient with right inferior eyelid swelling involving the conjunctiva and inferior transorbital approach through lower eyelid traverse incision was used and complete excision of lesion was done. Histopathology was suggestive of solitary plasmacytoma of the eye lid. Patient was referred to oncological unit for chemotherapy.

One patient with right orbital swelling and was approached by subfrontal craniotomy and complete excision of frontal sinus mucocele with duroplasty and exteriorization of frontal sinus done. This patient is fine and no postoperative complications.

One patient with huge tumour involving eye-nose-cranium (Figure 7) and was approached by frontoorbital craniotomy along with Weber-Fergusons approach and complete excision of the tumour with enucleation of eye with anterior cranial fossa base repair was done. Histopathology was suggestive of sinonasal-adenocarcinoma. Patient developed postoperative CSF rhinorrhoea which stopped with five days continuous lumbar CSF drain.

One patient who had long standing nonhealing ulcer over right fronto orbital region with complete destruction of eye (Figure 10) was approached by wide local excision of fronto-temporo-orbito-zygomatic bone along with enucleation of the eye and closure with local pedicle graft repair. Histopathology was suggestive of angiosarcoma. Unfortunately patient had prompt recurrence with in 1year.

One patient who presented with a huge mass involving the eye-nose-forehead (Figure 11) was approached by frontoorbital craniotomy along with Weber-Fergusons facial degloving with medial orbitotomy–ethmoidectomy along with enucleation of eye and duroplasty and anteriorcranial fossa base repair. Patient had immediate postoperative CSF leak, which stoped with lumbar CSF drain for 5 days. Histopathology and culture sensitivity was suggestive of mucormycosis. Patient was treated with intravenous liposomal Amphotericin-B for 14 days followed by oral Fluconazole for 14 days.

Two patients with posttraumatic orbital ateriovenous malformation following penetrating injuries involving the upper eyelids and both were operatred through superior transorbital approach and incision extended from medial to lateral canthus. First the feeding arterial supply were coagulated and the nidus was completely excised and venous were lastly coagulated. One patient required reconstruction of the eyelid with local pedicle graft.

One patient with orbitofrontal swelling (Figure 12) was operated by frontal craniotomy with excision of

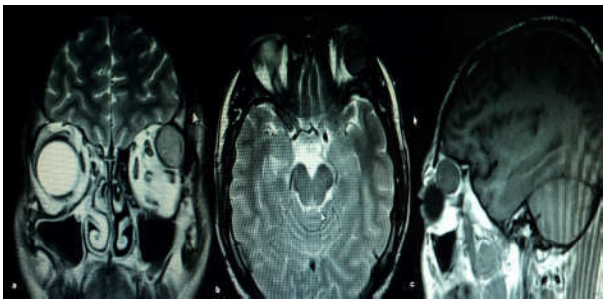
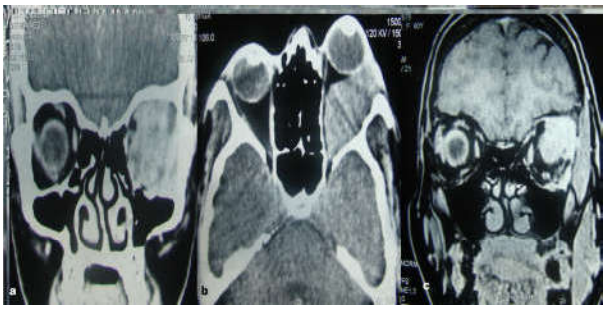


Fig. 1:



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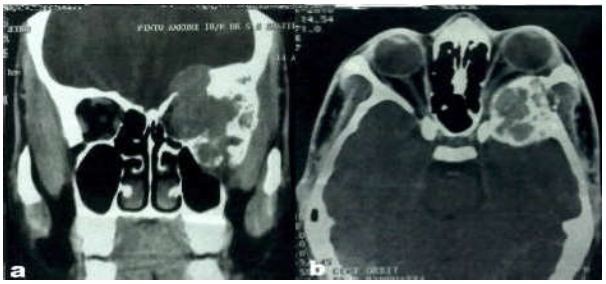

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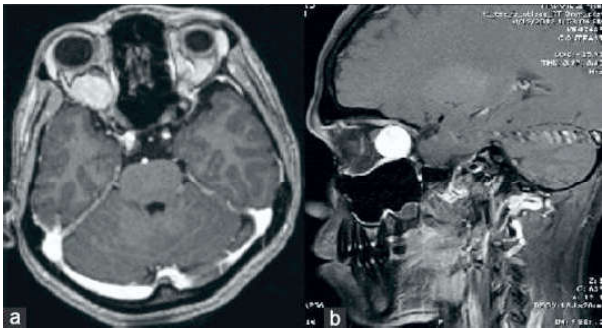

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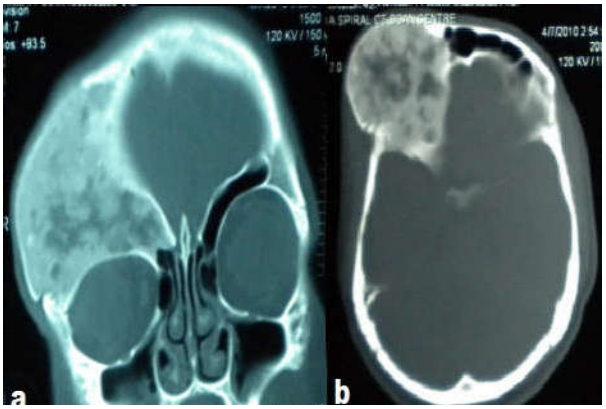

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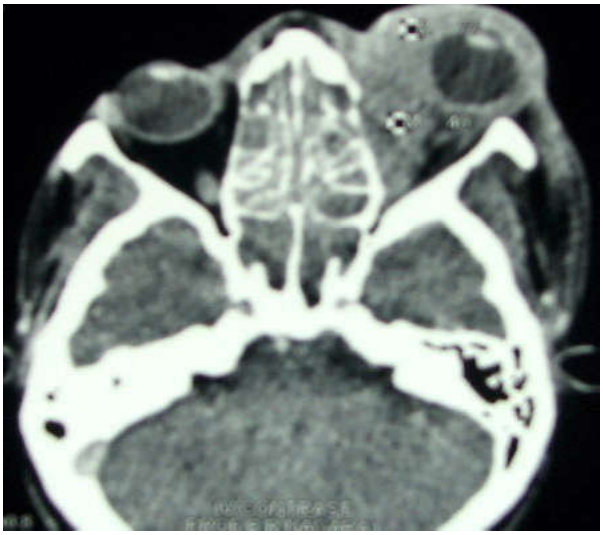

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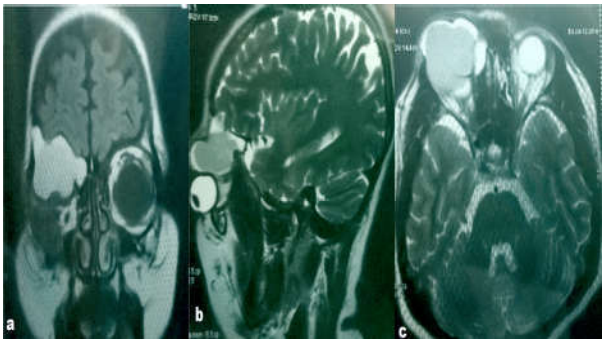

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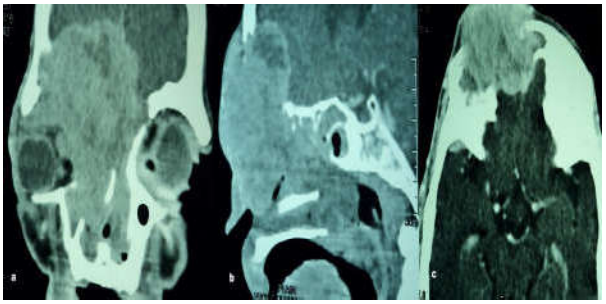

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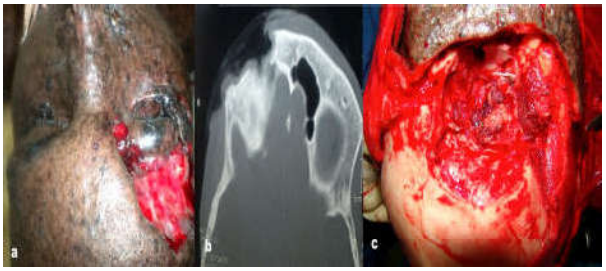

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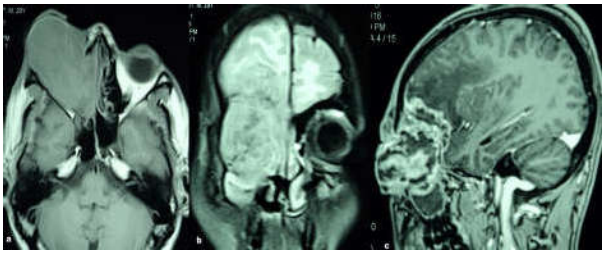

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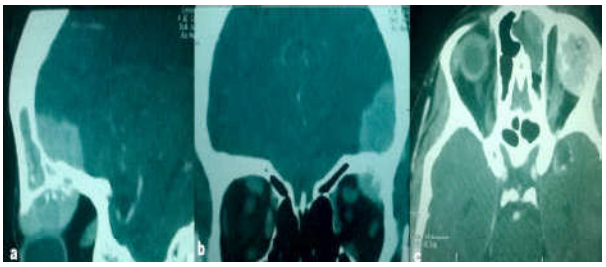

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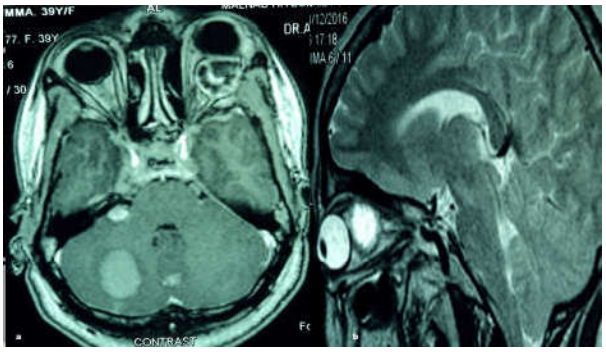

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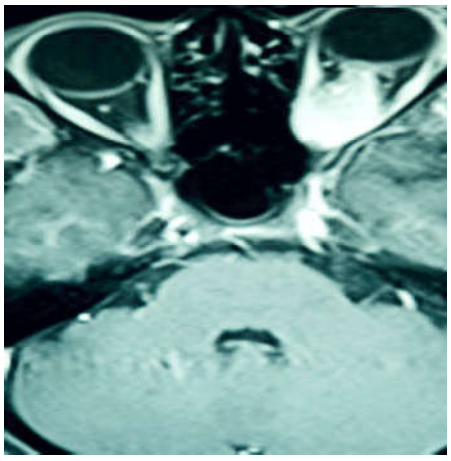

Fig. 13:

Table 1:

Diagnosis	No of Patients	Symptoms
Lacrimal gland adenoma	1	Proptosis & headache
Lacrimal adenoidcystic carcinoma	3	Pain, Proptosis & Visual loss
Recurrent maxillary adenocarcinoma with intraorbital extension	1	Facial numbness
Orbital apex meningioma	3	Proptosis &Diminution of vision
Sphenoid wing meningioma with intraorbital extension	1	Proptosis
Intraconal abducens nerve schwanoma	1	Pain
Intraorbital extraconal sensory nerve schwanoma	1	Pain & Proptosis
Plexiform neurofibrma with pthysis bulbi	1	Cosmetic disfiguration
Right frontoorbital Ewing's sarcoma	1	Proptosis
Right frontal solitary fibrous dysplasia	1	Proptosis
Left intraorbital lymphoma	1	Proptosis with loss of vision
Lower Eye lid solitary plasmacytoma	1	Painless Swelling
Frontal mucocele with intradural and orbital extension	1	Proptosis
Sinonasal carcinoma with intra orbital extension	1	Pain, Proptosis & Loss of vision
Fronto orbital angiosarcoma	1	Non healing ulcer & Pthysis bulbi
Sinoorbital mucormycosis with intracranial extension	1	Proptosis, pain & Pthysis bulbi Pain
Eyelid /orbital AVM	2	Pulsatile proptosis with conjunctival injection.
Metastases	1	Proptosis
Total patients	23	-

Table 2: Showing clinical profile of orbital tumors of series published by various authors.

Author and year	Patients (men/women)	Mean age year	Primary orbital tumors (%)	Secondary from contiguous structures (%)	Metastatic (%)	Proptosis (%)	diplopia	tightness/ pain/headache (%)	Decresed vision (blind eye)
Our series	23(12/11)	43	14(60.9%)	8(34.8)	1(4.3)	18	0	11	9(7)
Ohtsuka, K, 2005[2]	244(114/130)	48.7	213(89)	23(9)	8(2)	NA	NA	NA	NA
Nevo Margalit, 2007[6]	41(16/25)	42.2	5(12.2)	34(82.9)	2(4.9)	11(26.8)	2	7(17.1)	NA
Paluzzi A et al, 2015[17]	12(6/6)	39.5	11	-	1	4	2	4	5(NA)
Jian T et al, 2015[18]	21								
A. Chiriac et al, 2012[19]	57(28/29)	38.4	54	3	NA	39(68)	NA	15(26)	24(NA)

Table 3: Showing surgical approaches, extent of excision and complications

Authors, year	no	Approach	Extent of excision (%)	Enopthalmos	CSF leak	Follow up in months(mean)	Mortality related to surgery
Our series	23	Lateral orbitotomy- 1 Fronto-temporo-orbital craniotomy -2 Fronto-temporo-zygomatico-orbital -8 Weber-Fergusons -1 Superior transorbital -3 Fronto-orbital craniotomy -3 Medial orbitotomy-1 Inferior transorbital -1 Subfrontal craniotomy-1 Fronto-orbital+Weber-Fergusons -2	GTR 21 STR-2		2	2year and 6 months	0
Nevo Margalit, 2007[6]	41	Lateral orbitotomy 13. Frontal craniotomy+obitotomy 23.	GTR30(73.2) STR 5(12.2) PR 2(4.9)	1	1	1 to 38 (20)	0

		Combined Craniotomy+ Weber-Ferguson 1. Weber-Ferguson alone 4.	Biopsy only 4(9.8)				
Paluzzi A et al, 2015[17]		Frontal craniotomy-orbitotomy 3 Lateral orbitotomy 1 Medial orbitotomy 1 EEA 3 Combined Craniotomy + EEA 2 Combined medial transconjunctival & EEA 2	GTR 11 PR 1	NA	NA	NA	0
Jian T et al, 2015[18]	21	Coronal scalp + lateral orbitotomy 3 Coronal scalp 4 Frontal craniotomy 6 Pterional					
A. Chiriac et al, 2012[19]		Superior orbitotomy 19(33) Fronto-pterional 12(21) Fronto-orbital 6(10) Bicoronal craniotomy 8(14) Fronto-orbitotemporal 10(18) Transfacial 1(2) Transpalatal & Weber orbitotomy 1(2)	NA	NA	NA	7 to 62(17)	0

GTR (gross total resection– complete excision)
STR (subtotal resection- upto 2% of lesion left behind)
PR (partial resection- more than 3% residual lesion)
EEA- endoscopy endonasal approach

lateral half of superior orbital rim. Histopathology and detailed post-operative work-up was suggestive of metastasis secondary to ovarian neuroendocrinal carcinoma.

Discussion

Literature search (Table 2) shows the percentage of primary orbital tumours in adult population to be 11-89% [15-18], secondary orbital tumours from adjacent contiguous structures 3-82.9% and metastasis 1-4.9%. In our series, we had 14 primary orbital tumours, 8 secondary orbital tumours and 1 metastasis.

In order to choose an effective surgical approach, not only tumour dimensions and localization alone, but also and as importantly the benign versus malignant character of the lesion need to be considered to plan effective acces to orbit all along the 360 degree around the optic nerve plane. While benign tumours may be observed and assessed conservatively as long as the (bin) ocular function remains unaffected [16], malignant entities require an immediate and more radical treatment, which apart from surgical tumour removal by means of an orbitotomy shallneed adjuvant chemo- or radiotherapeutical measures.

The optic nerve meningioma, though benign, poses challenges with high probability of worsening of vision owning to interruption of the central retinal artery at its point of medial penetration of the optic nerve sheath whenever gross total excision is tried and partial excision however, allows the tumor to infiltrate the intraconal compartment upon regrowth, and in this matter [19] radiation therapy then becomes an important. We had similar situation that only partial excision was possible in one patient and when complete excision of the meningioma was done in two patients, both had 3rd nerve palsy and complete diminution of vision.

The surgical excision of large and massive orbital tumours carries significant risks of surgical complications including visual loss, exophthalmos, strabismus, and third nerve palsy [20]. These complications may occur as consequence of excessive traction, reduced working room, blind dissection behind the increased bulk of the tumour or release of tight adhesions between the tumour and the surrounding orbital structures.

Also there is need for reconstruction of orbit, skull base and give acceptable aesthetic look. Hence it is desirable to deal with these challenges by multidisciplinary team approach, undertaken by

well-experienced neurosurgical team along with ophthalmic/orbital team and plastic surgeon,while operating tumours involving the orbit-cranial junction, the superior orbital compartment to facilitate optimal removal of the tumour along with skull base sealing without compromising the good cosmetic results.

Conclusions

Orbit being a complex anatomical entity and management of complex tumours poses a challenging surgical problem. Although a significant percentage of these tumours are treated by the ophthalmologist alone, proficiency with a multitude of approaches and collaboration with a neurosurgeon is often required, especially for tumours that are located deep within the orbit, are large, or have an intracranial extension. Technical advances and modifications in surgical techniques along with involvement of plastic surgeons have decreased surgery-related morbidity and involvement of radiation oncologist to the team for adjuvant radiotherapy/chemotherapy would be beneficial for prolonged recurrence free and better quality of life.

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