Research Paper

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Case Report of Orbital Oncocystic Carcinoma

Medical Science

KEYWORDS : Oncocystic carcinoma, Proptosis, Periorbital swelling, FNAC

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ABSTRACT

OC is an unusual proliferation of cytomorphologically malignant oncocytes with adenocarcinomatous architecture phenotypes mainly found in glandular tissue. A report of a rare occurrence of an adult case of orbital oncocystic carcinoma. A 52 year old male type 2 uncontrolled diabetic, presented with right unilateral eccentric proptosis. Left eye was unremarkable. Fundus, Colour vision, and Intra ocular pressure was normal in both eyes. Ultrasonography showed grade 1 prostomegaly and hepatic stenosis. On Orbital CT, heterogeneously enhancing lesion at nasal and inferotemporal quadrants surrounding the right globe and extending to the apex of right orbit, with superolateral displacement of the globe was noted. Fine needle aspiration cytology report from the mass was suggestive of adenoid cystic carcinoma. Patient underwent right orbital exenteration and right partial maxillectomy under GA. Histopathological features were in favour of Oncocystic carcinoma.

Introduction

Oncocystic carcinoma (OC) is an unusual proliferation of cytomorphologically malignant oncocytes with adenocarcinomatous architecture phenotypes mainly found in glandular tissue [1, 2]. The terms oncocytic carcinoma, oncocytic adenocarcinoma, malignant oncocytoma and malignant oxyphilic adenoma are synonymous [3]. This tumor represents 5% of all oncocytic salivary gland neoplasms and less than 1% of all salivary gland tumors [4]. OC may occur in many sites in addition to the salivary glands, including the nasal and thoracic cavities, ovary, kidney, thyroid gland, breast and parathyroid. The majority of OC cases have occurred in the parotid glands [5].In this case report we describe oncocystic carcinoma in orbital cavity which is extremely rare.

Case report

A 52 years old male was examined in eye OPD with complaints of watering and swelling of right eye of 1.5 years duration. Patient is a known type 2 diabetic since 3 years under treatment with Metformin & Glibenclamide.

In observation eccentric proptosis RE was present. Best corrected visual acuity of right eye was 6/9 and 6/6 partial in left eye. Colour vision test with Ishihara plate reading: 13/13 (OD/OS).

Intra ocular pressure (Applanation tonometry) was within normal limit in both eyes. OD=16mmhg OS=16mmhg

Hertel's exophthalmoscopy: OD-24mm, OS-22mm

Pupils both eyes -normal

EOM –OD- showed restriction of movement in all directions.

Diplopia was present in dextroelevation.

Slit lamp examination showed conjuctival chemosis of right eye with lacrimation.

Dilated fundus examination: normal

Paraclinical evaluations

-Biochemistry reports showed High FBS & 2hpp (143-241)
-Haematology reports was within normal limits
-Ultrasound scan of abdomen, showed Grade 1 Prostomegaly and Hepatic stenosis

-CT scan of orbits showed:

Ill defined attenuation heterogeneously enhancing lesion at superior nasal and inferior quadrants of the right orbit, surrounding the right globe and extending to the apex of right orbit encasing the medial and inferior recti muscles. The right globe appeared displaced superolaterally, indenting of the right optic nerve was noted. [Fig 1-3]

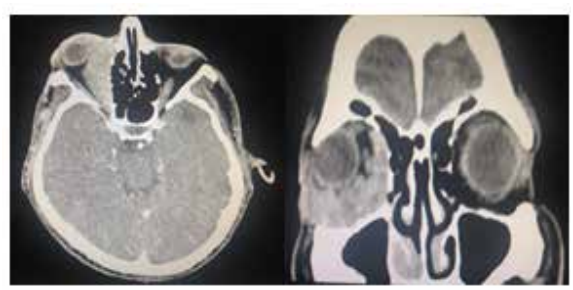

Fig1-Axial section **Fig2-Coronal section**

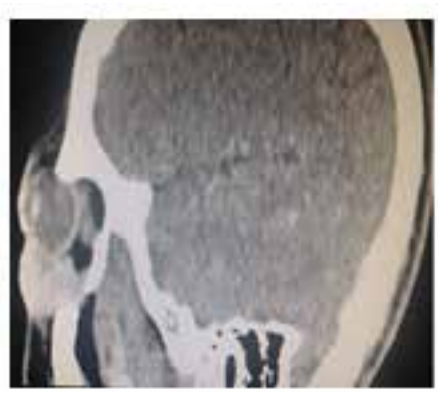

Fig3-Sajital section

FNAC (Fine needle aspiration cytology) from right orbital mass

Smears studies showed adequate cellularity against a haemorrhagic background. Tumor cells were arranged in small clusters, singles and also acinous pattern. Many large foamy mac-

rophages also seen. Cells are large with mild anisonucleosis and adequate eosinophilic cytoplasm. Occasional myxoid area noted. Dispersed plasmacytoid cells and mesenchymal material seen at few places .All features were suggestive of orbital malignancy, Adenoid cystic lesion.

-Procedure:
Patient [Fig 4] underwent right orbital exenteration [Fig 5] and right partial maxillectomy under GA.

Fig4

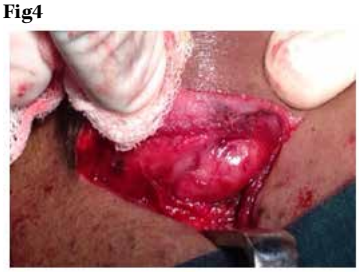

Fig5

-Histopathological features
of the mass were in favour of oncocystic carcinoma.

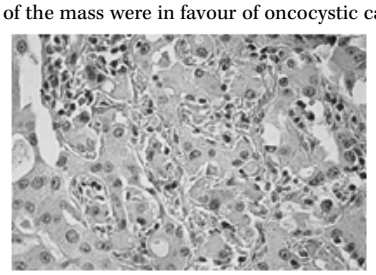

Fig 6

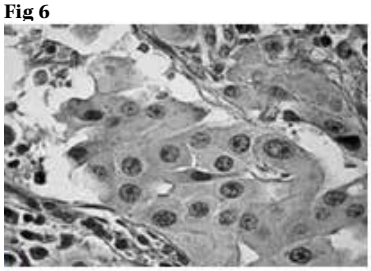

Fig 7
-Histopathological image showing peri-neural invasion by the tumor and cohesive clusters of neoplastic cells [Fig 6]

- High magnification image demonstrating oncocytic cells with abundant and finely granular cytoplasm and moderately pleomorphic nuclei located centrally or peripherally. [Fig 7]

Discussion
A variety of tumors and pseudotumors can involve the orbit. Study by Shield JA, Shields CL, Scartozzi R.(2004), showed in series of 1264 lesions 64% were benign and 36% were malignant.

The percentage of malignant tumors increased with age, with malignancies being common in older patients because of the higher incidence of lymphoma and metastasis in the elderly.

The most common orbital tumors in a clinical practice of ocular oncology include :lymphoid tumors (11%),idiopathic orbital inflammation (11%),cavernous haemangioma (6%),lymphangioma (4%),meningioma (4%),juvenile pilocytic astrocytoma (4%),metastatic breast cancer (4%),orbital extension of uveal melanoma (3%), capillary haemangioma(3%), rhabdomyosarcoma (3%), dermolipoma (3%), herniatedorbital fat (2%), dermoid cyst (2%), varix (2%), and dacryops [6]

OC is an extremely rare malignancy in orbit and prognosis is poor as it is a high-grade neoplasm with infiltrative growth pattern and tendency to recur and metastasize. OCA may involve the caruncle, the conjunctiva, the lacrimal sac, and more rarely, the lacrimal gland [7-10].

Treatment options are according to site of origin, histologic type or both and mainly are lesion resection with radiotherapy or combined radio/chemotherapy to prevent distant metastasis. Nakada et al. (1998) published a review of 28 cases of oncocytic carcinoma of the parotid gland [11]. They concluded that distant metastasis appeared to be the most important prognostic feature of oncocytic carcinoma; local lymph node metastasis was not necessarily a critical factor in the overall prognosis. Distant metastasis sites include the lung, liver, kidney, mediastinum, thyroid gland and bone.

Patients with malignant oncocytoma appear to have good short-term survival, but poor long-term survival [12]. The average survival period has been estimated at 3.8 years with metastasizing tumors [13].

This patient has had radiotherapy to prevent orbital recurrence following surgery and being followed up for metastasis.

Conclusion
Oncocytic carcinoma is an extremely rare malignancy in the orbital cavity. Standard treatment and prognosis is unclear. Prophylactic lesion dissection with radiotherapy or combined radio/chemotherapy in case of metastasis may be indicated for tumors larger than 2 cm in diameter as in the case reported. . Clinicians should perform a careful follow-up, as distant metastasis appeared to be the most important prognostic feature. Further investigation of the prognostic markers and correct treatment protocols, for patients with oncocytic carcinoma are required as more cases of OC are reported. The prognosis of oncocytic carcinomas is not well known because of their low incidence.

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