**How to Cite**:
Das, B. B., Vendhan, K. E., & Jeyaprakash, B. (2022). Topic-infraorbital xanthogranuloma associated with eccentric proptosis-based on surgical excision: A case report. *International Journal of Health Sciences*, *6*(S4), 4757–4761. https://doi.org/10.53730/ijhs.v6nS4.9246

# Topic-infraorbital xanthogranuloma associated with eccentric proptosis-based on surgical excision: A case report

**Dr. K. Ezhil Vendhan**
Professor, Department of Ophthalmology, Vinayaka mission's Kirupananda Variyar Medical College and Hospitals, Salem, Tamil Nadu. India

**Dr. B Jeyaprakash**
Associate professor, Department of Ophthalmology, Vinayaka mission's Kirupananda Variyar Medical College and Hospitals, Salem, Tamil Nadu, India

**Dr. Binayak Bibek Das***
Resident, Department of Ophthalmology, Vinayaka mission's Kirupananda Variyar Medical College and Hospitals, Salem, Tamil Nadu, India
*Corresponding author

***Abstract*---**Adult onset xanthogranuloma (AOX) is one of the very uncommon four varieties of adult xanthogranulomatous disease diagnosed mostly by characteristic histology. The work focusses on excision of the mass in the control of recurrence and associated periocular manifestations. The entity of above case report is very rare and unique and needs proper treatment and careful follow up to prevent cosmetic disfigurement.

***Keywords*---**adult onset xanthogranuloma, histocytes, proptosis, incision, excision biopsy.

## Introduction

AOX is one of the four uncommon entities of AXD confirmed mostly by histologic examination. It is a very rare group of entity with multiple organs like Heart, lungs, bone, retro-peritonium, orbit/ocular adnexa involvement. The symptoms vary depending on the part of the body involved to exophthalmos, proptosis, visual alteration, HTN, Heart and lung disease etc. Mostly has 4 varieties-Adult onset xanthogranuloma, Necrobiotic Xanthogranuloma, adult-onset asthma and peri-ocular xanthogranuloma and Erdheim Chester disease (ECD)[1]. Histopathologically, these varieties are mostly denoted by presence of "Hallmark cells" especially foamy histiocytes, Touton type giant cells, varying degree of

fibrosis. This study focuses on unilateral infra-orbital xanthogranuloma and associated recurrence and occurrence after FESS.

**Case report**

A 36-year-old male visited the opd with lobulated mass lesion in medial extraconal space of right orbit with peri-orbital swelling below right lower eyelid with partial decreased vision and restricted extra ocular movement on medial side, pain over swelling. The patient is a known case of undergoing microdebrider FESS twice, with ethmoidal polyp and fungal sinusitis in right eye. Patient is not known case of Diabetes mellitus, Hypertension, Asthma.

Figure 1: Patient picture

On MRI PNS and MRI Scan orbit, mass Effect is seen over optic nerve which is displaced laterally. Mass Effect was also seen over medial rectus muscle. Size of lesion is 6 cm*2.3cm*3.1cm. There is extension to medial aspect of pre-septal space, no intra-cranial extension. The mass is hyperintense in anterior part and hypointense on posterior part on T2. On MRI Brain, no significant abnormality was detected, mucosal thickening in right paranasal sinuses were seen. On incision biopsy, histiocytes, multinucleate giant cells, granulomatous inflammatory, dense collagenous cells were seen.

On Microscopy, extensive areas of necrosis were seen with rim of epitheloid granulomatous composed of epitheloid cells, multinucleate giant cells, lymphocytes, fibroblasts. Eccentric proptosis was noted from lateral orbital margin to apex of cornea and was noted to be 23mm.On palpation of orbital margin of lower lid, medial 1/3rd was obscured. Transillumination was absent. Naso-lacrimal test showed regurgitation of clear fluid from lower punctum.
On fundus examination, fundic reflex was dull, cup disc ratio within normal limit, background was normal.

**Differential Diagnosis**

Mucormycosis, Encephalocele, Hydatid cyst, Neurogenic tumor, lymphoma

**Examination and investigations**

**Systemic Examination**

CVS-S1 n S2 Heard
P/A -soft, NAD
R/S-B/L Air entry-Equal
CNS-conscious and oriented

**Local examination**

Examination of eye-periorbital swelling present in right eye
Examination of Ear-pre-auricular, pinna, post auricular region, external auditory canal, tympanic membrane, Middle ear mucosa was normal for both ears
Examination of nose-skin over root, bridge, dorsum of nose, columella, vestibule was normal
Right part of nose showed crust on anterior rhinoscopy
Examination of Throat
Oral cavity-teeth, lip, anterior 2/3 rd of tongue, hard and soft palate, buccal mucosa was normal
Examination of Oropharynx
Anterior pillar, tonsils, posterior pillar, post pharyngeal wall was normal

**Investigations**

Urea-20
Creatinine-0.6
HbsAg, HIV 1,2-negative
RBS-108 mg/dl
Hb-14. 2 mg/dl
BT-1 min 30 secs
CT-4 minutes
Pus culture and sensitivity-culture revealed contaminants

**Treatment**

Tab. Pan 40mg, tab Augmentin 625 mg, tab para 500 mg was given
Intralesional triamcinolone, Tab. Zerodol SP, refresh tears were given

**Course on Hospital**

Patient came with complaint of right infraorbital swelling. Patient started on antibiotics. Condition improved somehow, CT showed mass in orbital space, histology done to reveal Xanthogranuloma, excision done. Patient was discharged[2].

**Result**

After the incision biopsy, the yellow sample collected post-operatively, the most probable and likely diagnosis is Infra-orbital Xanthogranuloma.

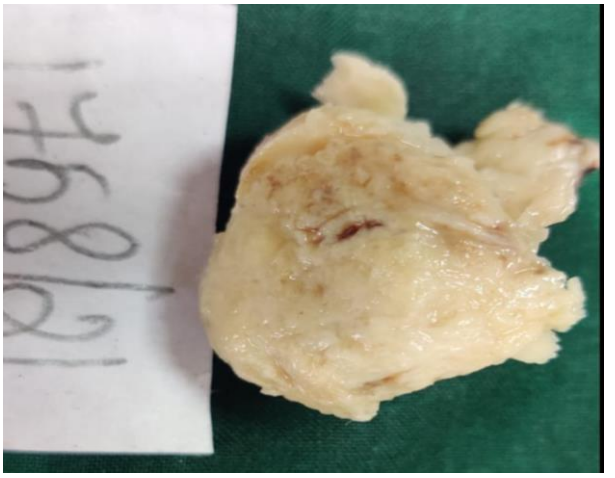

Figure 2: Excised xanthogranuloma mass yellowish

**Discussion**

Eyelid or ocular adnexa skin lesion can be presented in the 4 syndromes. In 1930,1st description of disease was done by William Chester and Jacob Erdheim. 4 types are discussed, AOX described as solitary lesion without systemic findings. Adult-onset asthma and peri-ocular xanthogranuloma (AAPOX) associated with immune dysfunction like asthma and lympho-proliferative disorders. Necrobiotic Xanthogranuloma is similar to AAPOX Erdheim-Chester disease (ECF) adds systemic involvement as pericardial or pleural effusions, retroperitoneal involvement, diabetic insipidus, Hepatosplenomegaly etc.

**Conclusion**

Xanthogranuloma is a very rare case study topic. The case has been studied due to its rarity and ophthalmic significance. Such cases are also important from cosmetic point of view to prevent recurrence and conserve the eyesight
Compliance with ethics Committee

**Conflict of interest**-No conflict of interest
**Funding**-Self funding by patient on insurance and scheme basis.

**References**

1. Chester W(1930) Uber lipidgranulomatose Virchow Archiv 279:561-602
2. Sivak-Callcot J. Rootman, J Rasmussen S et al(2006) Adult Xanthogranuloma disease of the orbit and ocular adnexa, new histochemical findings and clinical review. Br J ophthal 90:602-608.
3. Suryasa, I. W., Rodríguez-Gámez, M., & Koldoris, T. (2021). Get vaccinated when it is your turn and follow the local guidelines. *International Journal of Health Sciences*, *5*(3), x-xv. https://doi.org/10.53730/ijhs.v5n3.2938

4. Suryasa, I. W., Rodríguez-Gámez, M., & Koldoris, T. (2021). Health and treatment of diabetes mellitus. *International Journal of Health Sciences*, *5*(1), i-v. https://doi.org/10.53730/ijhs.v5n1.2864
5. Djuraev, A. M., Alpisbaev, K. S., & Tapilov, E. A. (2021). The choice of surgical tactics for the treatment of children with destructive pathological dislocation of the hip after hematogenous osteomyelitis. *International Journal of Health & Medical Sciences*, *5*(1), 15-20. https://doi.org/10.21744/ijhms.v5n1.1813