Case Report

A Rare Case of Intraocular Adipocytic Metaplasia in a Phthisical Eye

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ABSTRACT

Introduction: Phthisis bulbi is an end stage ocular disease caused by injury, long term eye disease and infection. The adipocytic metaplasia within the ocular coat is a rare variety of metaplasia.

Case: An 18 year-old female presented with complaints of severe dimness of vision in the right eye for last 11 years.

Observation: Slit lamp examination showed total cataract, shallow anterior chamber and enophthalmos in the right eye. As the right eye was blind, enucleation with silicon ball implant was advised. Histopathological examination of the eyeball showed evidence of phthisical eye, exudative retinal detachment with extensive osseous and adipocytic metaplasia.

Conclusion: Adipocytic metaplasia of retinal pigment epithelium is a very rare feature observed in intraocular pathology.

Key words: Adipocytes; intraocular; metaplasia; phthisis bulbi.

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INTRODUCTION

Phthisis bulbi is an end stage ocular disease caused by injury, long term eye disease and infection. The affected eye is shrunken and has little to no vision. The intraocular pressure in the affected eye is very low or non-existent. Disorganisation of intraocular structures, exudative retinal detachment (ERD) and ossification of intraocular lens are common histopathological features of phthisical eye (Tripathy et al., 2018).

The adipocytic metaplasia within the ocular coat is a rare variety of metaplasia. The retinal pigment epithelium (RPE) are considered to be pluripotent and have the capacity to differentiate into mesenchymal phenotype including adipocytes. Chronic inflammation, trauma or long standing retinal detachment can cause RPE to differentiate into mesenchymal cells resulting in intraocular adipocytes formation. It usually requires a couple of years for adipocytes to appear (Jakobiec et al., 2018). The pre-adipocytic factors viz. insulin-like growth factor-1, CCAAT-enhancer-binding protein, peroxisome proliferator-activated receptor- play a crucial role. The metaplastc adipocytes have high expression of lipogenic genes and production of adipokines like adiponectin, leptin and transforming growth factor- (Kumar et al., 2004; Schulz et al., 2009). Fluorescein dye staining can be used for diagnostic purpose in intraocular pathology (Das et al., 2020).

CASE REPORT

An 18-year-old female patient presented to a tertiary institute of northeast India with the chief complaint of severe dimness of vision in the right eye for 11 years which was gradual and worsening. There was associated intermittent throbbing pain in the right eye for two years. There was history of typhoid fever when she was seven year old and could not see anything by the right eye since that time. On examination, vision in the right eye was perception of light negative and left eye was 6/6. Slit lamp examination showed total cataract, shallow anterior chamber and enophthalmos in the right eye. Intraocular pressure was found to be very low (6 mm of Hg) in the right eye. Dilated fundus examination revealed no view in the right eye and the left eye was normal. B-scan ultrasound of the right eye showed decreased axial length, echogenic lens, high reflective echoes in the vitreous cavity. There was gross increase in retino-choroidal thickness with phthisical change in the right eye. Vitreo-retina clinic was consulted next. As the right eye was blind, enucleation with silicon ball implant was advised after patient and parental consent and it was carried out by oculoplasty surgeon. Enucleated eyeball was subjected for pathological examination in the ocular pathology laboratory.

Grossly, enucleated eyeball measured in all dimensions. Optic nerve was cut flush to the surface. Transillumination defect was not seen. Eyeball was sectioned vertically. The lens was calcified. (Figure 1). Microscopic examination of gross tissue material in fluorescein dye showed lobules of fat (Figure 2).

The haematoxylin-eosin stained tissue sections showed edema of the basal layer of the corneal epithelium. Anterior chamber was shallow with closure of the angle. The sclera was thickened throughout the eyeball. Choroid showed oedema, cystoids degeneration with sparse chronic inflammatory cells infiltration.

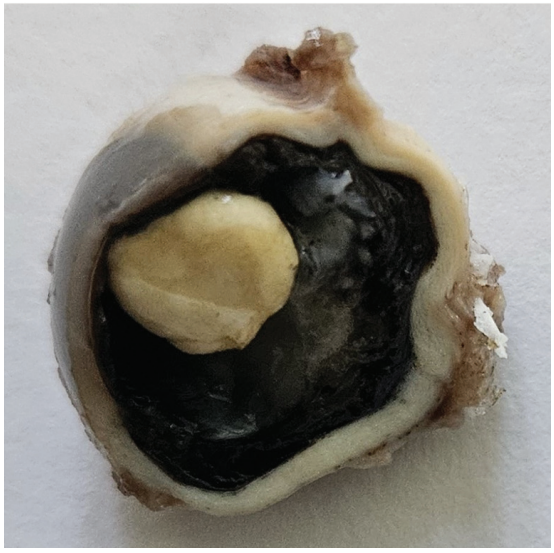

Figure 1: Gross enucleated eyeball showing calcification of intraocular lens.

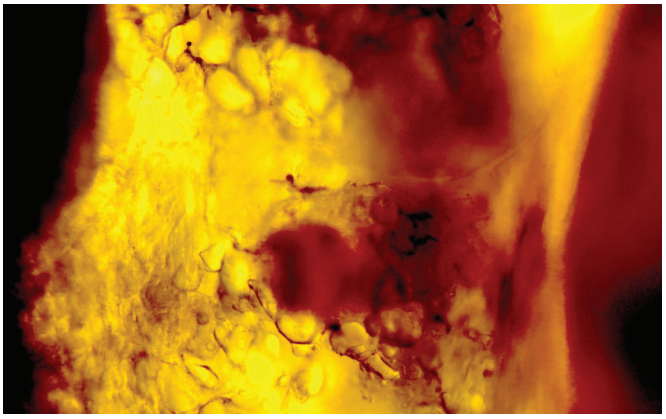

Figure 2: Fluorescein dyed photomicrograph of gross tissue material showing lobules of fat.

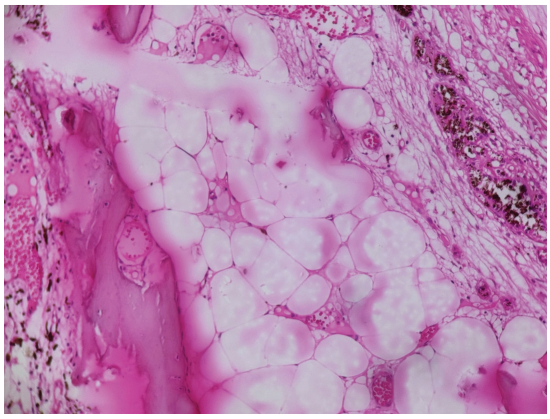

Figure 3: Presence of mature adipocytes inside the eyeball (haematoxylin and eosin stained photomicrograph, 400X).

Choriocapillaries were dilated and congested. The ERD noted with extensive osseous and adipocytic metaplasia (Figure 3). Hyaline degeneration was also noted at places. No tumour cells were identified. Intraocular lens was calcified.

DISCUSSION

Our case was very unique and very few such cases have been reported in the field of ophthalmic pathology till date. In a healthy eye, adipocytes are not typically present. This process may occur in various parts of the eye, including retina, ciliary body or vitreous. Sometimes intraocular adipose tissue can also be found in eyes with persistent hyperplastic primary vitreous (Font et al., 1969). Epithelial-mesenchymal transition of intraocular lens is another contributing factor of adipocytic metaplasia. Inflammatory cytokines play a crucial role in stimulating this

cellular transformation. Multipotent stem cells of RPE are a subpopulation of adult human RPE cells that can be stimulated in the lab to self-renew and differentiate into stable RPE cells, mesenchymal cells and neural cells (Jakobiec et al., 2018).

CONCLUSION

The intraocular adipocytic metaplasia in phthisis bulbi symbolises the future role of retinal pigment epithelium as a source of multipotent stem cells which has many therapeutic implications in order to salvage the vision of pathological eye. Yet another potential application is to awaken dormant endogenous repair.

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