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Bilateral Isolated Ankyloblepheron Filiforme Adnatum – A Case Report

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Abstract
Ankyloblepheron filiforme adnatum(AFA) is a rare congenital anomaly of the eyelids which is usually isolated but can be associated with other systemic abnormalities. Timely separation of the lids is important to prevent amblyopia. We report a case of a 7 years old boy with bilateral AFA with no systemic associations from North Eastern India which was surgically corrected.
Keywords: *ankyloblepheron filiforme adnatum, congenital anomaly*

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I. Introduction

Ankyloblepheron filiforme adnatum (AFA) is a rare congenital anomaly of the eyelids comprising single or multiple fine bands of extensile tissue connecting the upper and lower lid margins at the grey line. It is usually isolated but can be associated with other rare systemic abnormalities

II. Case Report

A 7 year old boy was brought to the Ophthalmology department of a tertiary hospital in the North Eastern part of India by his mother with the complaints of inability to open both his eyes completely since birth. The child had a normal vaginal delivery at fullterm with an uneventful antenatal period. There was no significant family history of any skin condition, cleft lip, cleft palate or similar eye condition.

On ocular examination, there were 2 bands of tissue with cilia in each eye near the lateral canthus connecting the upper and lower eyelids with normal ocular movements and lacrimal passage. The vertical palpebral height of the patient was 6mm in both the eyes. The medial and lateral bands of tissues on each eye measured around 3mm$_x$3mm and 0.5mm$_x$2mm respectively. The unaided visual acuity of the patient was 6/6.

Figure 1: Two bands of connective tissue bridging the lower and upper lids near the lateral canthus of the right eye

Figure2: Two bands of connective tissue bridging the lower and upper lids near the lateral canthus of the left eye

Referral was done to the paediatrics, ENT, orthopaedics, dental and dermatology departments for complete systemic examination to rule out any underlying syndrome. A diagnosis of bilateral Ankyloblepheron filiforme adnatum without any systemic association was made. A simple excision of the bands from the lid margins was done under general anaesthesia. Light cauterization was done at the bleeding points.The procedure was uneventful. Detailed anterior segment and posterior segment examinations was done on the post operative day one which revealed normal findings. A normal intraocular pressure of 13mmHg and 15 mm was noted on the right and left eye respectively. The child was discharged with the plan of genetic testing.

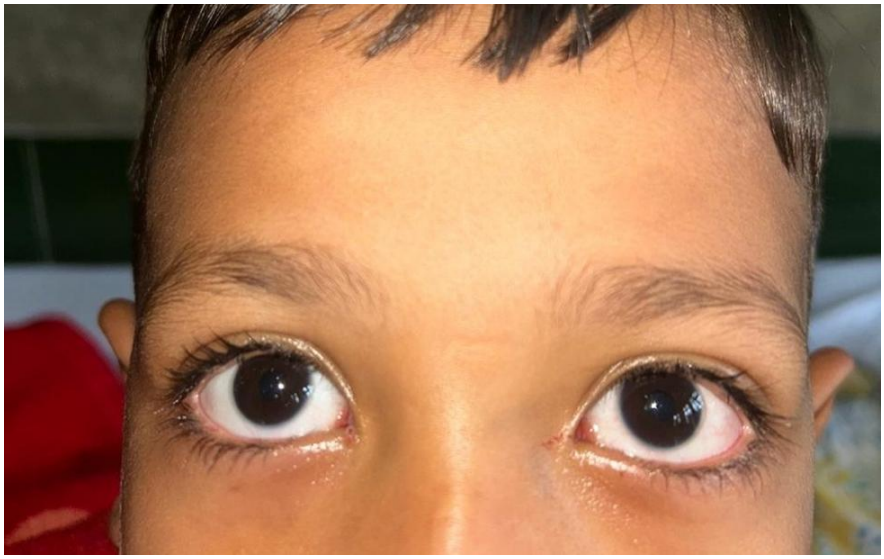

Figure 3: Post operative day 1 picture of the patient

III. Discussion

AFA is a rare congenital anomaly first described by Von Hasner in the year 1881[1].During the development of eyelid structures, the epithelial fusion of the eyelid margins occur at around 9th week of gestation. The lid margins usually remain fused until about the end of the 5th fetal month. Complete separation usually does not occur until about the 7th fetal month.[2]. When the lid margins fail to separate at birth, it leads to ankyloblepharon though the exact etiology is unknown. The currently accepted theory is that this condition is due to temporary epithelial arrest and rapid mesenchymal proliferation, allowing union of eyelids at abnormal positions[3]. AFA was divided into 4 subgroups by Rosenman *et al*. According to which group I and II are sporadically inherited, and the remaining groups, group III and IV have autosomal dominant inheritance pattern. Group I have no associated abnormalities, group II is associated with cardiac or central nervous system abnormalities, group III is associated with ectodermal syndrome while group IV is associated with cleft lip or cleft palate[4]. Bacal *et al* suggested a fifth group: AFA in association with chromosomal abnormalities.

A significant central ankyloblepharon may interfere with vision and cause amblyopia whereas medial or lateral ankyloblepharon may restrict the visual fields. A medial ankyloblepharon may also obstruct the punctum.

Sporadic cases have been described in the literature however it can be associated with wide range of systemic conditions which includes the ACE or Hay Wells syndrome, Edward syndrome, CHANDS , Popliteal pterygium syndrome, cardiac anomalies, Rapp-Hodgkin syndrome etc . Some of these multisystemic conditions share mutation in the p63 genes.p63 is a member of the transcription factor family and does not represent a clasic tumor suppressor gene as p53.p63 is rather a key regulator in limb, epithelial, and craniofacial development[5]. Mohan et al reported one patient with Hay –Wells syndrome developed end staged renal failure secondary to reflux neuropathy related to urinary tract abnormality[6]. Scott MH, Richard J M, Farris BK also reported case of AFA associated with infantile glaucoma and iridodonesis[7].

Definitive management of ankyloblepharon is surgery. But multideciplinary approach for early detection and intervention is needed not only to avoid the development of amblyopia but also to exclude associated systemic malformations and the complications. Our case, fortunately did not have any sign of systemic involvement nor ambylopia.

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