A Rare Case of Ankyloblepharon Filiforme Adnatum

Rajesh Powar¹, Nilesh Kumar¹, Arvind Y. Yakkundi², Niharika²
¹Department of Plastic Surgery, JN Medical College, Belagavi, Karnataka, India
²Department of Ophthalmology, JN Medical College, Belagavi, Karnataka, India

Ankyloblepharon Filiforme Adnatum (AFA) is an extremely rare congenital anomaly occurring due to failure of separation of eyelids during fetal development. Despite its dramatic presentation, the treatment is bedside release of the adhesions and thus restoring vision and cosmesis. The hesitation on the part of treating physician or pediatrician can lead the child to amblyopia, and we report this case to add to the existing literature for a better understanding of the disease.

Keywords: ankyloblepharon filiforme adnatum, eyelids

Introduction
Ankylolblepharon Filiforme Adnatum (AFA) is a rare congenital anomaly characterized by the presence of dense adhesion bands between the upper and lower eyelids which may be partial or complete, and may or may not be associated with other systemic findings or syndromes.¹,²

Case Report
A female new-born, born out of non-consanguineous marriage to a 23-year-old primigravida at 38 weeks of gestation by a cesarean section in view of meconium-stained liquor, was referred to the authors with complaints of inability to open the eyelids due to the presence of skin adhesions by a neonatologist at 6 hours of life. The mother had developed gestational diabetes during the pregnancy, and the antenatal period was uneventful. There was a similar history of inability to open the eyelids in her paternal great-grandfather which was left untreated. The child was 2950 gms at birth and no immediate post-natal complications were found. Ocular examination revealed bilateral multiple dense connective tissues between the upper and lower eyelids with the inability to fully separate the lids (Figure 1). The cornea and sclera below it seemed to be normal though detailed examination was not possible. Systemic examination was unremarkable and no syndromic features were found on meticulous head-to-toe examination. Bedside release of the bands was performed using tenotomy scissors without any use of anesthetic or sedative agents.

Physical restraint was used to stabilize the child’s head to avoid any unwanted injury to the underlying globe. Post-release, the ocular examination was normal bilaterally. The bands were found to be attached to the grey line suggested by the presence of raw area after the release (Figure 2 & 3). She was having normal blink reflex, eyelid movements and extra-ocular muscle movements bilaterally. In view of the exposed area at the release site, Moxifloxacin eye ointment (0.5%) was prescribed twice daily to be applied at the lid margins. No patching or taping was done.

Discussion
The first report of AFA dates back to 1881 when it was described by Josef von Hasner to be characterized by partial or complete full thickness adhesion of upper and lower eyelids at the ciliary edges. It was usually found to be an isolated finding of sporadic origin then; but an autosomal dominant pattern was found frequently when it occurred in conjunction...
with other syndromes like AECS: Ankyloblepharon Ectodermal defects-Cleft lip/palate Syndrome, also known as Hay-Wells syndrome.3 Other frequent associations found are Infantile Glaucoma, Edwards' syndrome, CHANDS i.e. curly hair, ankyloblepharon and nail dysplasia syndrome.4 In view of such associations, Rosenman in 1980 divided AFA into 4 subgroups which were:

- Isolated (Group 1);
- AFA associated with cardiac or central nervous system anomalies (Group 2);
- AFA associated with ectodermal syndromes (Group 3); and
- AFA associated with cleft lip and/or palate (Group 4).2

Recently, a 5th group has been proposed for cases associated with chromosomal abnormalities.5 The case being reported here thus was found to be of Group 1 i.e. Isolated AFA. The pathogenesis of AFA has not been clear, though the failure of apoptosis at the 5th month of gestation when the upper and lower eyelids separate has been postulated.6,7 Temporary epithelial arrest with rapid mesenchymal proliferation has also been thought to be one of the pathogenic mechanisms.8 Treatment of AFA is the simple release of the bands with the help of scissors, which can be done bedside. If being performed by beginners, mild sedation is thought preferable to negate the chances of sudden movement of the head by the child which may cause globe injury. No topical anesthesia or local infiltration is indicated. Patching or taping should be avoided as there are chances of the raw area to adhere to each other. Spontaneous movement of eyelids; if left unpatched, will help to prevent such secondary ankyloblepharon. Antibiotic ointments should be applied, lid hygiene should be maintained. Follow-up is usually not required but can be warranted in view of other associated syndromes, and thus varies from case to case. Spontaneous lysis of such bands seems to be a far-fetched idea and delayed mechanical release on account of hesitancy of the treating doctor can cause the child to develop unwarranted amblyopia and cosmetic problems. Though release at a later stage of life can normalize the appearance, the amblyopia if set in, will cause a major socio-economic impact on the life of the individual.

**Conclusion**

The objective of this report was to emphasize on the simple treatment of a very rare condition which can prevent significant vision loss that may result due to delay in treatment. Also, a detailed examination to rule out syndromic association in cases of AFA is strongly recommended.

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**Address for correspondence**

Nilesh Kumar
Doms
Resident,
Department of Ophthalmology,
JN Medical College, Belagavi,
Karnataka, India
Email id: n.nilesh.kumar@gmail.com

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