

Ankyloblepharon filiforme adnatum- a case report & review of literature

Sharma R.¹, Sami I.², Sharma N.³, Singh S.⁴, Ranjan R.⁵

¹Dr. Reena Sharma, Assistant Professor, ²Dr. Ifsa Sami, Senior Resident, ³Dr. Namita Sharma, Senior Resident, ⁴Dr. Sonali Singh, Senior Resident, ⁵Dr. Ravi Ranjan, Professor; all authors are attached with Department of Ophthalmology, UPUMS, Saifai (U.P.) India.

Corresponding Author: Dr. Ifsa Sami, Senior Resident, Ophthalmology, UPUMS, Saifai (U.P.) India. E-mail: ifshika20@gmail.com

Abstract

Ankyloblepharon filiforme adnatum (AFA) is a rare congenital anomaly characterized by presence of bands between eyelids. It can present as an isolated finding or as a part of syndrome. A 2-month-old male was presented to us with multiple fine bands of skin in both the eyes. Rest examination was normal. Excision of these bands was done with satisfactory result. We also reviewed the literature for the disease and its association. Although rare, AFA can cause stimulus deprivation amblyopia if not treated in time. Thorough examination should be performed in such cases to rule out other systemic and ocular associations.

Keywords: Amblyopia, Ankyloblepharon filiforme adnatum, Congenital abnormalities, Stratified squamous epithelium.

Introduction

Ankyloblepharon is the fusion of part or all of the eyelid margins. Ankyloblepharon filiforme adnatum (AFA) a variant of ankyloblepharon, was first described by Josef von Hasner in 1881 [1]. It is a rare congenital abnormality in which single or multiple strands of fine connective tissue join the upper and lower lids [2] at

any site arising from the 'grey line', anterior to the meibomian gland orifices and posterior to the cilia [3]. AFA may have sporadic occurrence as in our case and an incidence of 4.4 per 100,000 births has been reported [4, 5].

Case Report

A 2 months old male was brought to the hospital by his parents with the complaint of inability to completely open the eyes. He was the first child born out of a non consanguineous marriage, normal antenatal and perinatal history and the mother had not taken any drugs or been exposed to radiation. Family history was negative for any ocular or birth defects. The examination showed a single filamentous band between the two lid margins in the right eye and three bands in the left eye [Figure 1].



Figure-1: Photograph showing picture of child at time of presentation showing AFA

Manuscript received: 20th May 2019

Reviewed: 30th May 2019

Author Corrected: 6th June 2019

Accepted for Publication: 11th June 2019

Case Report

The bands were of varied width and were situated between the cilia and meibomian gland orifice causing inability to fully separate the lids. Rest anterior segment examination was normal. The systemic examination showed no abnormal features. Surgery was planned and the base of the bands were excised under general anaesthesia at the lid margins with minimal bleeding, without any intra and post operative complications. Post operatively, he had normal blink reflex, unrestricted eyelid movement with normal dimensions of palpebral fissure. Patient was followed up for two years without recurrence [Figure 2].



Figure 2: Photograph showing picture of child at two-year follow-up

Discussion

AFA may present as an isolated anomaly [6] such as in our patient or in association with the wide spectrum of congenital abnormalities including cleft palate and hare lip [7, 8] hydrocephalus, meningomyelocele, and imperforate anus [9] bilateral syndactyly [10], patent ductus arteriosus [11], ventricular septal defect [12] and in association with various syndromes like Edwards' syndrome¹³, Hay-Wells syndrome (a variant of the ectodactyly-ectodermal dysplasia-cleft lip palate syndrome) [14], the popliteal pterygium syndrome [15] etc. It usually presents in both the eyes but may be unilateral rarely [5, 16]. Its ocular association includes iridogoniodysgenesis and glaucoma [17].

Normally during the early period of gestation eyelids remain fused and get separated completely till the seventh month [18]. When it fails to do so, AFA develops. Various theories have been given in the past on the etiology of AFA, but failure of apoptosis at a critical stage in eyelid development has been suggested as the main factor [19].

Histology of the AFA demonstrated that the strands consist of vascularized central core surrounded by stratified squamous epithelium [20]. Considering the classification given by Rosenman et al, the present case belongs to its type 1 subgroup [7]. These bands cannot be lysed spontaneously. Though surgery at a later stage can solve the cosmetic issues, but amblyopia cannot be reversed. Diagnosis of AFA is mainly clinical and the treatment of choice is surgical resection of the fibrous bands either under intravenous sedation [21] or under topical anesthesia [22]. Some even suggested resection

without sedation or local anaesthesia [5]. Whatever might be the mode of anaesthesia, prompt surgical correction should be done to avoid development of amblyopia.

The major practical importance of this anomaly is that when it occurs, it should alarm the physician to the possible presence of other congenital problems that may account for high mortality and morbidity in future.

Funding: Nil, **Conflict of interest:** Nil

Permission from IRB: Yes

References

1. Von Hasner. Ankyloblepharon filiforme adnatum. Z Heilkd. 1881;2:429.
2. Duke-Elder S. System of ophthalmology. London-Kimpton, 1964;3:869-871.
3. Khanna VN. Ankyloblepharon filiforme adnatum. Am J Ophthalmol. 1957;43(5):774-777. doi: [https://doi.org/10.1016/0002-9394\(57\)91522-2](https://doi.org/10.1016/0002-9394(57)91522-2)
4. Cizmecci M, Kanburoglu M, Akelma AZ, Tatli MM. A stitched eye in the newborn: ankyloblepharon filiforme adnatum. J Pediatr. 2013;162(1):211-212. doi: [10.1016/j.jpeds.2012.08.041](https://doi.org/10.1016/j.jpeds.2012.08.041). Epub 2012 Oct 10.
5. Gruener AM, Mehat MS. A newborn with ankyloblepharon filiforme adnatum: a case report. Cases J. 2009; 2(1):8146. doi: [10.1186/1757-1626-0002-0000008146](https://doi.org/10.1186/1757-1626-0002-0000008146).

Case Report

6. Sarowa S, Jain K, Agarwal R. A newborn with ankyloblepharon filiforme adnatum. *J Clin Ophthalmol Res* 2018;6(2):63-65. doi: 10.4103/jcor.jcor_46_17
7. Rosenman Y, Ronen S, Eidelman AI, Schimmel MS. Ankyloblepharon filiforme adnatum: congenital eyelid-band syndromes. *Am J Dis Child*. 1980;134(8):751-753.
8. Reddy MP, Raghu SR. Congenital fusion of jaw and ankyloblepharon filiforme adnatum: malformation and multiple systems anomaly. *Indian J Plast Surg*. 2012; 45(3):557-559. doi: 10.4103/0970-0358.105977.
9. Kazarian EL, Goldstein P. Ankyloblepharon filiforme adnatum with hydrocephalus, meningomyelocele, and imperforate anus. *Am J Ophthalmol*. 1977;84(3):355-357. doi: [https://doi.org/10.1016/0002-9394\(77\)90679-1](https://doi.org/10.1016/0002-9394(77)90679-1)
10. Akkermans CH, Stern LM. Ankyloblepharon filiforme adnatum. *Br J Ophthalmol*. 1979;63(2):129-131. doi:10.1136/bjo.63.2.129
11. Lobstein A, Haarscher A. Congenital filiform ankyloblepharon and cardiac malformation. *Bull Soc Ophthalmol Fr*. 1953;2:128-130.
12. Zhu L, Ju R. Bilateral ankyloblepharon filiforme adnatum associated with cardiac defects: A case report. *AMJ* 2018;11(1):34-36. doi: 10.21767/AMJ.2017.3297
13. Williams MA, White ST, McGinnity G. Ankyloblepharon filiforme adnatum. *Arch Dis Child*. 2007; 92(1):73-74. doi:10.1136/adc.2006.103069
14. Hay RJ, Wells RS. The syndrome of ankyloblepharon, ectodermal defects and cleft lip and palate: an autosomal dominant condition. *Br J Dermatol*. 1976;94(3):277-289. doi: <https://doi.org/10.1111/j.1365-2133.1976.tb04384.x>
15. Gorlin RJ, Sedano HO, Cervenka J. Popliteal pterygium syndrome. A syndrome comprising cleft lip-palate, popliteal and intercrural pterygia, digital and genital anomalies. *Pediatr*. 1968;41(2):503-509.
16. Elbasher SE, Zakharchenko L, Boyle MA. Ankyloblepharon filiforme adnatum. *Arch Dis Child Fetal Neonatal Ed*. 2018;103(3):F207. doi: 10.1136/archdischild-2017-313932. Epub 2018 Jan 5.
17. Scott MH, Richard JM, Farris BK. Ankyloblepharon filiforme adnatum associated with infantile glaucoma and iridogoniodysgenesis. *J Pediatr Ophthalmol Strabismus*. 1994;31(2):93-95.
18. Sharkey D, Marlow N, Stokes J. Ankyloblepharon filiforme adnatum. *J Pediatr*. 2008; 152 (4):594. doi: 10.1016/j.jpeds.2007.12.051.
19. Mohamed YH, Gong H, Amemiya T. Role of apoptosis in eyelid development. *Exp Eye Res*. 2003; 76(1): 115-23. doi: [https://doi.org/10.1016/S0014-4835\(02\)00269-5](https://doi.org/10.1016/S0014-4835(02)00269-5)
20. Gupta SP, Saxena RC. Ankyloblepharon filiforme adnatum. *J All India Ophthalmol Soc*. 1962; 10: 19-21.
21. Özyazgan I, Eskitasçoglu T, Dündar M, Karac S. Hereditary isolated ankyloblepharon filiforme adnatum. *Plast Reconstr Surg*. 2005;115(1): 363-364. doi: 10.1097/01.PRS.0000146092.43354.4B
22. Ioannides A, Georgakarakos ND. Management of ankyloblepharon filiforme adnatum. *Eye (Lond)*. 2011; 25(6): 823. doi: 10.1038/eye.2011.26. Epub 2011 Feb 25.

.....

How to cite this article?

Sharma R, Sami I, Sharma N, Singh S., Ranjan R. Ankyloblepharon filiforme adnatum- a case report & review of literature. *Trop J Ophthalmol Otolaryngol*. 2019;4(4):303-305. doi: 10.17511/jooo.2019.i04.08

.....