

Ankyloblepharon Filiforme Adnatum: A Case Report

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Abstract: Introduction: Ankyloblepharon filiform adnatum (AFA) is defined by partial or complete fusion of the eyelids by webs of skin. It can be interest a part of the eyelid or its totality. Diagnosis should be done at an early stage of life. It is basically surgical. Case information: We describe a male new born at his twentieth day of life was referred for assessment of his both right and left eyelids. His was born on term. Pregnancy and delivery went normally. However there were first degree consanguinity. The baby was healthy apart from multiples bands of tissue between upper and lower eyelids of both of his eyes. These bands were axial, covering the pupil, and prevented full opening of both eyelids. The baby underwent surgery consisting of excising the bands of tissue with scissors at the level of each eyelid margin. Eye examination did not reveal any abnormality. Two months later, the follow up showed no anomaly. Conclusion: Ankyloblepharon filiform adnatum may be rare but it is potentially amblyogenic. However, the major practical importance of this anomaly is, perhaps, to alert the physician as to the presence of other associated congenital anomalies and the treatment should be performed as soon as the diagnosis is done.

Keywords: Ankyloblepharon Filiforme Adnatum, Congenital, Eyelid, Surgery

1. Introduction

Ankyloblepharon filiform adnatum is characterized by partial or complete adhesion of the ciliary edges of the upper and lower eyelids at the gray line, by single gold multiple fine bands of extensible tissue, which reduced the palpebral fissure by interfering with the movement of the lids. It is amblyogenic and its association with other congenital may account for high mortality and morbidity [1]. We report here a case of Ankyloblepharon filiform adnatum in a male newborn and describe its management.

2. Clinical Case

A male new born at his twentieth day of life was referred for assessment of his both right and left eyelids. His was born on term. Pregnancy and delivery went normally. The antenatal, intranatal, and postnatal periods were uneventful. The mother denied taking any drugs except iron and vitamin supplements. There was no history of any X-ray exposure. There was no family history of congenital anomalies,

however there were first degree consanguinity. The baby was healthy apart from multiples bands of tissue between upper and lower eyelids of both of his eyes. These bands were axial, covering the pupil, and prevented full opening of both eyelids (figure 1). Apart from this, he was perfectly healthy without any other congenital abnormalities identified after a thorough pediatric assessment.



Figure 1. (Before treatment) Multiple fibrous bands connect the upper and lower eye lids and prevent opening of the eyes.

The baby underwent surgery consisting of excising the bands of tissue with scissors at the level of each eyelid margin. Eye examination did not reveal any abnormality. Two months later, the follow up showed no anomaly (figure 2).



Figure 2. (After treatment) Appearance of the eyes after the division of the band tissue.

3. Discussion

First described by Von Hasner in 1881, Ankyloblepharon filiform adnatum is a rare benign congenital anomaly, usually associated with multiple and complex malformations, suggestive of an autosomal dominant inheritance of varying degrees of penetrance. Sporadic cases have, however, also been described [2]. No sex affinity has been discussed to date [3]. The fusion of eyelids is a normal stage in human development. The developing eyelid margins remain fused until the fifth gestational month but may take up to the seventh month of gestation to be completely separated [4]. Fusion seen at birth as in Ankyloblepharon filiform adnatum is abnormal.

The etiology of Ankyloblepharon Filiform Adnatum is unknown, but failure of apoptosis at a critical stage in eyelid development has been suggested. Usually, Ankyloblepharon Filiform Adnatum constitutes a solitary malformation, as in our case, with sporadic occurrence. However, it can be associated with several disorders [5].

Diagnosis of ankyloblepharon is based on clinical features. It is defined by partial or complete fusion of the eyelid margins [6]. A significant central ankyloblepharon may interfere with vision and cause amblyopia in congenital variety.

The length of the bands varies from 1 to 10 mm and breadth varies from 0.3 to 0.5 mm and are invariably extensible. The bands always lie between the cilia and orifices of the tarsal gland. The band is always composed of a central vascular connective tissue strand surrounded by pavement epithelium. The connective tissue is usually highly cellular and embryogenic in nature, but Cordero found muscle fibers and numerous sub-epithelial glands in the histological study of the band. When the bands are cut, bleeding may occur [1]. Pathogenesis of the condition is disputed. Various theories include a simple defect in separation, pathologic growth of the skin of inflammatory origin, an epithelial defect in fetal life through which connective tissue grows, most likely due to trauma, for example, fingernail trauma in utero. But the most accepted theory is that of pure aberrance of development, due either to a temporary arrest of the growth of epithelium or more probably, an abnormally rapid proliferation of mesoderm allowing union at certain points of the mesenchyme of the lid folds without epithelial interposition [1].

It is usually a solitary malformation of sporadic occurrence, but can occur in an autosomal dominant pattern associated with cleft lip and palate in most familial cases. In

some patients, it appears as a part of Edward's syndrome (Trisomy 18), Hay-Wells syndrome (a variant of ectodactyl-ectodermal dysplasia-cleft lip-palate syndrome), popliteal pterygium syndrome (characterized by intercrural webbing of the lower limbs), and curly air-ankyloblepharon-nail dysplasia (CHANDS). It may also be reported in association with hydrocephalus, meningocele, imperforate anus, bilateral syndactyly, infantile glaucoma, and cardiac problems such as persistent ductus arteriosus and ventricular septal defect [7-10].

An unusual report of a multiple malformation syndrome with Ankyloblepharon filiform adnatum, with cleft lip and palate, bilateral popliteal pterygia, bilateral complete syndactyly of 2nd and 3rd toes and hypoplastic nails, accessory nipple, and partially descended testis has also been reported [2].

Management of ankyloblepharon is mainly surgical. Detailed systemic assessment by an experienced pediatrician is essential to rule out coexisting pathology and proper management of congenital ankyloblepharon. Fine bands of adhesions can be broken by forcibly separating the lids or with use of muscle hook, sharp scissors or a scalpel [11]. The fine remnants at the lid margin usually shrink and resolve. Bipolar cautery forceps can be used at the bases of fine filaments to release the adhesions. In congenital ankyloblepharon, visual prognosis is usually good if timely intervention is performed. Cosmetic appearance after separation of the bands is usually very good [12]. A spontaneous resorption of the bridles can be observed after a few months. The prognosis depends on the associated anomalies [13-17].

Moreover, considering the pattern of inheritance, genetic counseling of the parents of the affected offspring may be suggested. However, the major practical importance of this anomaly is, perhaps, to alert the physician as to the presence of other associated congenital anomalies [18].

4. Conclusion

Ankyloblepharon filiform adnatum may be rare but it is potentially amblyogenic and it requires further investigation. Treatment should be performed as soon as the diagnosis is done.

Conflicts of Interest

All the authors do not have any possible conflicts of interest.

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