Case Study

Ankyloblepharon filiforme adnatum in a neonate

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INTRODUCTION

Ankyloblepharon Filiforme Adnatum (AFA) describes a rare congenital condition characterized by a partial or complete fusion of upper and lower eyelids unilaterally or bilaterally by single or multiple bands (1). AFA may be a sporadic occurrence or associated with other congenital defects, few of which may be life-threatening and require prompt treatment (2). It can easily be treated surgically (3). If not diagnosed and treated early, a significant central AFA can lead to occlusion amblyopia (4).

CASE REPORT

A one-day-old female neonate was brought to our clinic for right eyelid evaluation. She was a symmetrically small-for-dates neonate with a birth weight of 2kg, second born of a non-consanguineous marriage to healthy parents. There was no history of any infection, radiation exposure, or teratogenic drug consumption during pregnancy. There was no family or sibling history of any significant ophthalmic or systemic illness. On examination, there was a partial fusion of the right eyelids with a band (Figure 1).

Figure 1: ***showing partial fusion of the right eyelids by a band of tissue***

The neonate could not fully open the right eye. By separating the eyelids, the opening could almost be doubled, proving that the bands were extensible. The left eye was normal. There were no systemic congenital anomalies clinically. Ultrasound scans of the abdomen and cranium and echocardiography were normal. The band was divided by one cut with Wescott conjunctival scissors under local anaesthesia. Tobramycin (0.3% w/w) eye ointment was applied to the site postoperatively. The patient was able to fully open the right eye after the procedure (Figure 2).

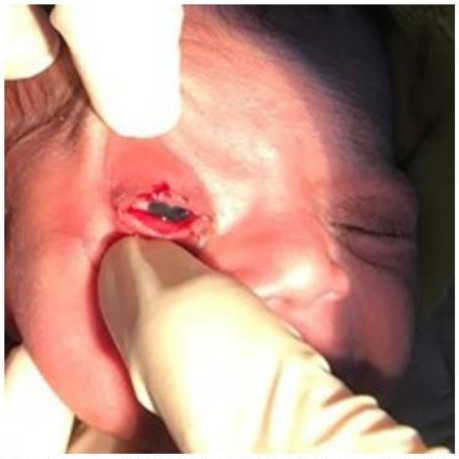

Figure 2: ***Immediately after the division of band***

There was point bleeding, which stopped without any major intervention. A detailed ocular examination, including fundoscopy, done the next day did not reveal any other additional anomalies. There were no systemic or ocular problems at two months of age (Figure 3).

Figure 3: ***Follow up at two months of age***

DISCUSSION

The word 'ankyloblepharon' is derived from the words "ankylos", which means restriction of movements, and "blepharon", meaning eyelids (5). Adhesions between the eyelids could be (6)congenital or acquired (7). AFA is a congenital variant characterized by a partial or complete fusion of the upper and lower eyelid ciliary edges with fine extensile band tissue (1). An Austrian Ophthalmologist, Josef Von Hasner, first reported it in 1881(1). It is a rare congenital malformation with an annual incidence of approximately 4.4 per 100,000 births (8). The fusion of upper and lower eyelids is a normal phenomenon until the 5th month of gestation, but they are entirely separated by the 7th month due to apoptosis. If the apoptosis fails, then the eyelid margins fail to separate and causes AFA (9).

The bands are single or multiple, unilateral or bilateral, lateral or central, and rarely medial (10). The band begins from the gray line between the meibomian gland orifices and cilia (11). It consists of central vascular connective tissue surrounded by squamous epithelium on histological examination(10). It may be an isolated anomaly as in our case (12) or may present with other abnormalities (13). The ophthalmic anomalies described are iridogoniodysgenesis and juvenile glaucoma (6).The reported systemic associations are Hay-Wells syndrome, trisomy 18, popliteal-pterygium syndrome, Curly Hair-Ankyloblepharon-Nail Dysplasia (CHAND) syndrome, cleft lip and palate, meningocoele, hydrocephalus, bilateral syndactyly, imperforate anus and cardiac conditions such as ventricular septal defect, patent ductus arteriosus (2) and atrial septal defect(14).

AFA was initially divided into four subgroups, of which the first two were sporadic groups, whereas the last two were autosomal dominant with variable expressivity(15). A fifth group was later described in association with chromosomal abnormalities by Bacal DA, *et al* (16) (Table 1). Our case belongs to Group I.

Table 1: ***Classification of AFA by associated congenital anomalies***

Group	Associated anomalies
I	None
II	Cardiovascular or central nervous system defects
III	Ectodermal syndrome
IV	Cleft lip and or cleft palate
V	Chromosomal anomalies

A significant centrally located band may cause occlusion amblyopia by interfering with vision, while bands on either side may restrict visual fields (17). It can easily be treated by releasing the bands with a blade (18) or scissors (19). Antibiotic ointment should be applied post-procedure and left unpatched as the eyelid's spontaneous movement will help prevent secondary ankyloblepharon (20). A detailed ophthalmic assessment, including a fundus examination, should be done post-procedure. This report stresses early identification and simplicity in treating this rare entity, leading to severe visual impairment if not treated in time.

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