Case Report

Ankyloblephron Filiform Adnatum: A Case Report

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Abstract

Ankyloblephron filiform adnatum (AFA) is a rare benign congenital anomaly that can arise either in isolation or associated with a syndrome. It should be treated as early as possible due to its ambylogenic potential. We report a case of a successfully managed newborn that had sporadic AFA detected at birth. Our case is unique in the sense that sporadic AFA with Atrial septal defect has not been reported in the literature.

Key words: Amblyopia, Ankyloblephron filiform adnatum, cardiac defects

Introduction

Ankyloblephron filiform adnatum is a benign rare congenital anomaly characterized by single or multiple bands of delicate connective tissue extending between the upper and lower lid margin leading to partial/full thickness fusion of the lid margins. This tissue begins from the grey line, and is located anterior to the Meibomian gland orifices and posterior to cilia (Alami et al. 2013). The eyelid margins typically remain fused until the fifth gestational month, but is an abnormal occurrence at birth (Greuner et al. 2009).

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CASE

A two-day-old female, born at term by Caesarean section, weighing 3.2 kg, to a multigravida mother was referred to ophthalmology for lack of spontaneous eye-opening. On examination, multiple bands of tissue were seen connecting the upper and lower eyelids. Eyelid opening was impaired, and anterior segment structures could not be discerned due to small eye-opening.

The antenatal history was unremarkable. The mother denied taking any medications during her pregnancy. The child was a product of a non-consanguineous marriage, and the family history was unremarkable. The siblings were normal. The baby was noticed as having bilaterally fused eyelids at birth. On examination, multiple bands of tissue connecting both the upper and lower eyelids with difficult eye-opening were seen (Figure1).

Systemic examination failed to identify any other congenital abnormalities except for an ejection systolic murmur (grade II/VI) best heard over the pulmonary area on auscultation. Routine laboratory tests were normal. Echocardiography examination revealed an atrial septal defect of size 2mm with predominantly left to right shunt.

The child was planned for early surgery under sedation because of amblyogenic potential. The bands of tissue in eyelids were initially crushed with artery forceps and then, excised with Vanna's scissors flush to the eyelid margin. The procedure was associated with minimal bleeding. Subsequent eye examination, including anterior segment, ocular motility and fundus, was normal. Figure 2 illustrates the picture of the patient on the 2nd postoperative day.

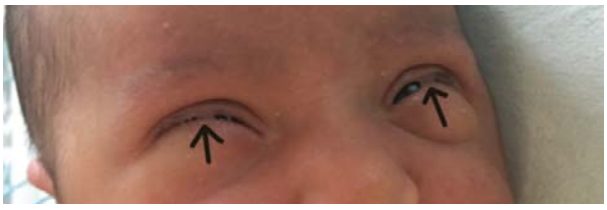

Figure 1: Photograph of baby showing bands of tissue connecting both lids (arrows).

Figure 2: Photograph after excision of the bands

Discussion

Von Hasner in1881 described Ankyloblephron filiforme adnatum (AFA) as a rare congenital abnormality of the eyelids. The condition is characterized by single or multiple bands of elastic tissue joining the upper and the lower eyelid margin. The etiology is mostly unknown, but most commonly postulated theory is that it results due to an interplay of temporary epithelial cell arrest and rapid mesenchyme proliferation leading to the union of lids (Alami B et al. 2013).

Treatment is aimed to eliminate the ambylogenic risk to the developing visual system. This report is presented because of the rare association of AFA with Atrial septal defect.

AFA usually presents as an isolated malformation occurring sporadically, or it can also occur in association with cleft lip and palate as an autosomal dominant inheritance.

It has also been reported with Edward syndrome, popliteal pterygium syndrome (webbing of the knees), Hay-wells syndrome (ankyloblepharon-ectodermal dysplasia clefting syndrome), and CHANDS (curly hair ankyloblepharon-nail dysplasia syndrome). Cardiac disorders such as ventricular septal defect and patent ductus arteriosus have been reported by Patil BB et al. (2001), but its association with the atrial septal defect has not yet been reported in the literature.

Detailed systemic assessment by an experienced pediatrician to rule out associated systemic abnormalities and timely surgical intervention to prevent amblyopia is, therefore, mandatory in the management of AFA. AFA was classified into four groups by Rosenman et al. (1980), and later, a fifth group was suggested by Bacal et al. (1993): AFA in association with chromosomal abnormalities. Our case fits in group 2 of the Rosenman classification.

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