Case Report

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An Approach to a Case of Ankyloblepharon

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Abstract

Ankyloblepharon is a rare congenital abnormality consisting of partial or complete adhesion of the upper and lower eyelids by single or multiple bands. Ankyloblepharon is potentially amblyogenic condition. It is generally isolated but can present with systemic diseases. Its presence should alert the need for a detailed systemic evaluation. We present a 1 month old baby who presented to our institute with bilateral adhesions of the upper and lower eyelids, and then patient is evaluated systemically and then surgery was planned. The adhesion was excised with the use of Radio-Frequency cauteryundershort sedation. The separation of the lidsbefore the development of visual milestones is potentially important to prevent the onset of stimulus deprivation amblyopia.

Keywords: Ankyloblepharon; Congenital abnormality; Eyelid bands; Amblyopia.

INTRODUCTION

Ankyloblepharon is partial or complete adhesion of the eyelids by webs of skin. It may be present since birth (congenital) or may be acquired.[1] Ankyloblepharon may be complete, partial or interrupted. The interrupted form is also known as ankyloblepharon filiforme adnatum (AFA). It was first described by Von Hasner in 1881. It causes decrease in the palpebral fissure height and limited movement of eyelids. It is a benign, rare but potentially amblyopic congenital malformation.[2] During embryonic life the eyelids stay fused until the 5th month of fetal life. At this period, they start to separate, but it remain partially attached until the 7th month.[2] This abnormality happens when the eyelids fail to separate, either partial or complete, resulting in shortening of the palpebral fissure. These limit eyelid movement. Ankyloblepharon may present as an isolated finding, may be associated with other congenital anomalies like hydrocephalus, meningocele, an imperforate anus, bilateral syndactyly, infantile glaucoma and cardiac problems such as patent ductus arteriosus and ventricular septal defects or it may be a part of a well defined syndromelike trisomy 18 (Edwards´ syndrome)[6], Hay-Wells syndrome (a variant of the ectodactyly-ectodermal dysplasia cleft lip palate syndrome)[7,8] the popliteal pterygium syndrome (characterized by intercrural

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webbing of the lower limbs), and CHANDS (curly haira-nkyloblepharon-nail dysplasia syndrome).[3] Therefore, it is important to look for systemic abnormalities.[3] Treatment of Ankyloblepharon is onlyremoval of tissue band from lid margins by simple excision.

CASE REPORT

A 1 month old male infant was presented to our OPD for the assessment of his eyelids of both eye to Sarojini Naidu Children Hospital, Prayagraj, UP, India. He was born at term, weighing 3.6kg. The baby was born at 39 weeks' gestation by normal vaginal delivery to a 27-year-old primi gravid female and 33-year-old healthy male, out of non-consanguineous marriage. The pregnancy was normal and uneventful, with no history of fever, rashes, drug intake or radiation exposure. There was no family history of congenital anomalies. A detailed systemic assessment did not reveal any other systemic congenital anomaly. Ocular examination showed a peripheral band arising from gray lines leading to adherence eyelids (Fig. 1), measuring about 0.5mm breath and 3mm in length.

Fig. 1: Pre-operative

The band of tissue was excised with Radio-Frequency cauteryin sterile conditions under mild sedation (Fig. 2).

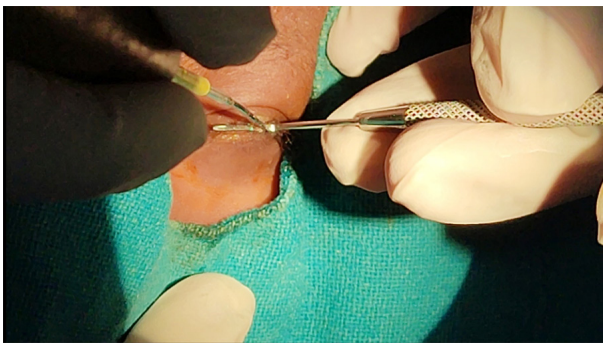

Fig. 2: Intra-Operative cutting of band using rf cautery

Chloramphenicol and Polymixin ointment two times daily for one week was prescribed. The posterior surface of the eyelids, ocular surface, ocular motility, anterior segment and fundus were normal. Intraocular pressures after excision was normal (Digitally). Post-operatively, on day 1 both eyelids were fully opened with no abnormality (Fig. 3). Pediatrician's review of the child indicated no other congenital abnormality, hence the diagnosis of an isolated ankyloblepharon was made.

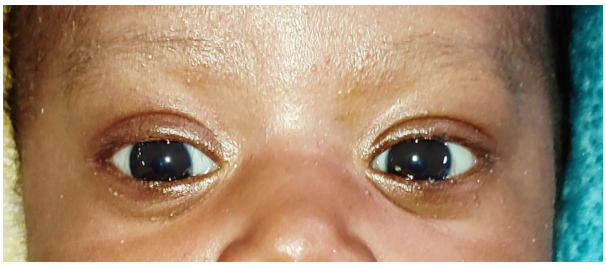

Fig. 3: Post-operative

DISCUSSION

Ankyloblepharon is a rare benign congenital anomaly, first described by Von Hasner in 1881. Adherance of the lid margins is a normal process in human development, but not a normal condition at birth. The eyelid margins in fetal life remain fused till the 5th month of intrauterine life, but sometimes it may not be completely separated till the 7th month. Ankyloblepharon presents as a single or multiple bands of tissue adhering the eyelids either unilaterally or bilaterally.[4] It may present as an congenital defect not associated with any systemic condition such as in our patient. Yet, it becomes important to actively look for any associated pathology. The ocular association of it is iridogoniodysgenesis with juvenile glaucoma.[5] It has been reported in the context of trisomy 18 (Edwards´ syndrome)[6], Hay-Wells syndrome (a variant of the ectodactyly-ectodermal dysplasia-cleft lip palate syndrome)[7,8], the popliteal pterygium syndrome (characterized by intercrural webbing of the lower limbs), and CHANDS (curly hair-ankyloblepharon-nail dysplasia syndrome). Other associations may include hydrocephalus, meningocele, an imperforate anus, bilateral syndactyly, infantile glaucoma and cardiac problems such as patent ductus arteriosus and ventricular septal defects.[9] Detailed systemic assessment by an experienced pediatrician is therefore imperative in the management of AFA. Rosenman *et al*[10,11] divide AFA into four subgroups (1. isolated; 2. associated with cardiac or central nervous system anomalies; 3. associated with ectodermal syndromes; 4. associated with cleft lip and/or palate) and indicated that groups 1 and 2

were sporadic and groups 3 and 4 were autosomal dominant with variable expressivity. Bacal *et al*[6] suggested a fifth group i.e. in association with chromosomal abnormalities. The aetiology of this abnormality is unknown and a number of theories have been proposed. The currently accepted theory is that this condition is due to an interplay of temporary epithelial arrest and rapid mesenchymal proliferation, allowing union of lids at abnormal positions.[8] This case report demonstrates the simplicity (by using Radio-Frequency Cautery) in treating Ankyloblepharon. Surgical correction should be performed immediately to reduce any risk of amblyopia, and enable full examination of the eye. It also alerts that its presence should alarm the clinician for the possibility of an underlying congenital disorder.

CONCLUSION

Ankyloblepharon is a potentially amblyogenic congenital abnormality of the eyelids and it also highlights that its presence should alert the clinician to the possibility of an underlying congenital disorder. This case demonstrate the simplicity (by using RF cautery) in treating Ankyloblepharon is quick, safe and prevents the risk of stimulus deprivation amblyopia.

Declaration of Patient Consent

Obtained all patient consent forms. The patient(s) attendant has given their written consent for their pictures and other clinical information to be reported in the journal.

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Conflicts of Interest: No conflicts of interest.

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