

# ANKYLOBLEPHARON FILIFORME ADNATUM IN AN AFRICAN BABY-A Case Report

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## Summary

Ankyloblepharon filiforme adnatum is a congenital malformation of the lid margins, wherein the lids are connected by fine strands of extensible tissue. It can occur sporadically or be inherited as an autosomal dominant gene with variable penetrance. The abnormality is easily amenable to surgical lysis, but it is important to examine the baby for other congenital abnormalities. We present a two-week-old baby with this condition, who like other associated ankyloblepharon-ectodermal dysplasia clefting syndrome (AEC) has severe skin desquamation.

**Key words:** congenital anomaly, congenital ankyloblepharon, lid malformation

## INTRODUCTION

The lid buds start growing in front of the developing eye at six weeks of intrauterine life (IUL). The two lids meet, fuse at eight weeks IUL and then separate during the fifth month.<sup>1</sup> The failure of the lids to separate is known as congenital ankyloblepharon. In true ankyloblepharon, the two lids are connected by webs of skin, either in part or throughout their length, thereby shortening the palpebral aperture. When the two lids are connected by fine extensile tissue, the terminology proposed by Von Hasner<sup>1</sup> is ankyloblepharon filiforme adnatum. Ankyloblepharon and ankyloblepharon filiforme adnatum often coexist with other congenital abnormalities grouped together as ankyloblepharon-ectodermal dysplasia clefting syndrome (AEC).<sup>2</sup> Ankyloblepharon filiforme adnatum can exist sporadically or as an autosomal dominant gene with variable phenotypic expression. We present a baby with

this syndrome which, to the best of our knowledge, has not been reported in blacks.

## CASE REPORT

A two-week-old baby girl was referred to Olabisi Onabanjo University Teaching Hospital with a history of inability to open both eyes since birth, and a fever for one week. She was seen in the neonatal ward and referred to the ophthalmologist after treatment for sepsis. The baby was a product of a full-term pregnancy. Antenatal history and delivery were uneventful. She is the fourth child in a family of four children. The parents are from the Edo ethnic group of Nigeria. The mother is 35 years old, while the father is 45 years. They both engage in fishing as a means of livelihood. The other siblings, two males and one female are normal. There was no family history of lid malformation, nor was there any history of other congenital abnormality. There was also no history of consanguinity. The mother denied ingestion of any drugs during pregnancy.

On examination, the baby weighed 3.5kg. She had patches of desquamated skin all over her body, including the forehead. The upper and lower lids of both eyes were connected by multiple fine strands of extensible tissue below the margin of the cilia. It was difficult to see the right globe, but the left globe was partially visible and was normal. Paediatric examination did not reveal any other congenital anomaly. Lysis of the strands was done with a scissors, under sedation with 2.5mg intramuscular diazepam. Bleeding was minimal and oculentum chloramphenicol was applied. Both globes were found to be normal after lysis.

## COMMENT

The term ankyloblepharon filiforme adnatum was first proposed by Von Hasner in 1881. It is a benign congenital anomaly which can be unilateral or bilateral.

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This baby had the bilateral type. Although the condition is benign it can coexist with other congenital anomalies such as cleft lip and palate,<sup>3</sup> severe skin erosions,<sup>4</sup> neurological abnormality and imperforate anus.<sup>5</sup> Patil<sup>6</sup> cited a case by Lobstein et al.<sup>7</sup> in which there was



cardiac abnormality. Roseman et al.<sup>8</sup> have divided ankyloblepharon filiforme adnatum into four subgroups. In subgroup 1 there is no associated abnormality. In subgroup 2 there is cardiac anomaly. In subgroup 3, there is ectodermal syndrome, and subgroup 4 is associated with a cleft lip and/or palate. He also concluded that subgroups 1 and 2 occur sporadically, while subgroups 3 and 4 were autosomal dominant with variable expression.

It would appear that our patient is in Roseman's subgroup 3, although there was no family history of a similar condition. Our patient had severe skin erosions with secondary infection similar to the cases reported by Vanderhoof.<sup>4</sup>

The globe in ankyloblepharon filiforme adnatum is often normal, although Scot et al.<sup>9</sup> have reported a case with infantile glaucoma and iridodysgenesis. Although, ankyloblepharon filiforme adnatum is a benign congenital abnormality, it may co-exist with other more serious conditions. It is therefore necessary to conduct a thorough examination of the baby.

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