

Case Report
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Congenital Monocular Deficit of Elevation and Myopia Case Report

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ABSTRACT

We present a case of congenital restrictive strabismus and axial myopia that was associated with an anomalous orbital structure identified by MRI and CT. This fibrous band was observed and resected during surgery.

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Case Report

Male, 11 years old who presented strabismus since the first year of life, his mother reported that it has worsened over time. He has mild developmental delay. His medical report referred anisometropic amblyopia of left eye. The cycloplegic measures showed in right eye (RE) was -0.75 D. In left eye (LE) was -15 D. Visual acuity with optical correction was 20/20 and 20/200 respectively. He presented monocular deficit of elevation, with ptosis and pseudoptosis, Bells phenomenon absent in LE and abduction deficit. In primary position the LE had 15 PD of Esotropia and 30 PD of Hypotropia, measured by Krimsky Test. The ocular motility presented marked limitation in upgaze and left gaze. Esotropia worsened in downgaze and left gaze due to limitation in the abduction of the left eye of -3 degrees. Hypotropia worsened in upgaze and left gaze (Figure 1).



Figure 1

Ptosis and pseudoptosis of left eye was observed. Bell phenomenon was absent in LE.

Orbital images (MRI and CT scans) showed superior displacement of the left medial rectus (LMR), slight inferior displacement of left lateral rectus (LLR) and slight nasal displacement of left inferior rectus (LIR) (Figure 2). In an CT scan, a structure can be identified that could correspond to a vestige of the retractor bulbi muscle (Figure 3). Examination under anesthesia: FDT was positive to elevation and abduction (Figure 4). The left eye was in ET and Hypotropia (Figure 5).

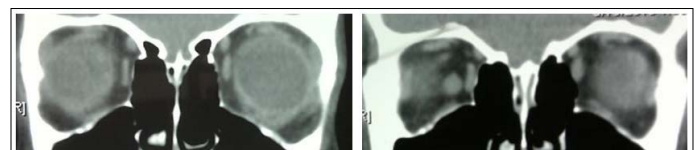
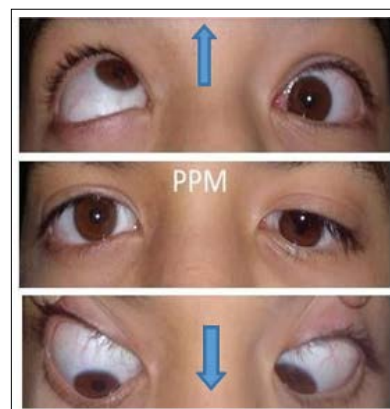
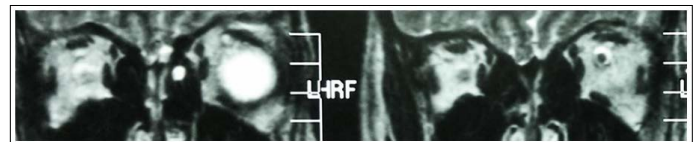


Figure 2



Figure 3



Figure 8



Figure 4

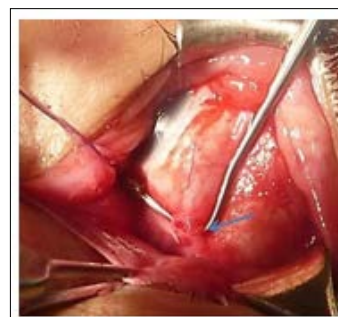


Figure 9



Figure 5

After the conjunctival incision was made, the rectus muscles are looked for. The LLR is displaced downwards (Figure 6). The LSR was hooked and partial loop myopexia was done between both without scleral passage according to the Yokoyama technique (Figure 7). The LMR was hooked and it was found elevated. It reached the midline making traction with the hook. Once it was uninserted the FDT became negative to abduction but remained positive to elevation, so the lower sector was carefully explored with a strabismus hook, to avoid the LIR and LIO (Figure 8). A tight fibrous band was located between the LMR and the LIR, which indent the sclera, making very difficult the scleral passage of the hook. After the section of it, the FDT normalized towards elevation. A 4 mm LMR recess with descens of half insertion was performed (Figure 9).

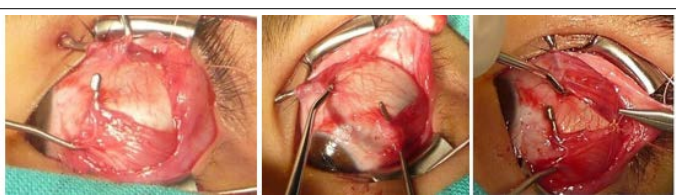


Figure 6,7

Results

The limitation in the vertical plane persisted after the surgery, an improvement was observed in the horizontal plane with XT of 12 PD in Primary position (Figure 10).

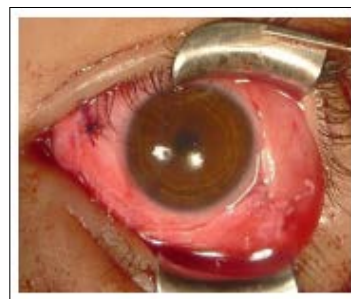


Figure 10



Evolution

It was indicated to continue treatment for amblyopia and use the lenses permanently.

The patient attended one month after surgery and did not return for subsequent check-ups. The need for Reoperation had been raised.

Discussion

The association of axial myopia and the presence of an abnormal orbital band has not been reported in the literature. It seems to be an uncommon case of restrictive strabismus with a double pathogenic mechanism. Lueder states that anomalous orbital structures (such as muscular or fibrous bands) that attach to the globe in rare instances may produce a mechanical restriction, resulting in incomitant motility disorders [1,2]. Of the 3 types of anomalous orbital structures described in the literature, one involves accessory muscle fibers that are innervated by the III and/or VI cranial nerves originating in the posterior orbit and inserting on the globe, optic nerve, or extraocular muscles [1-4]. Fibrous or muscular bands may be located beneath the ERMs; they also may originate from the ERMs themselves and insert in abnormal locations or may occur as discrete anomalous muscles that take origin in the posterior orbit and insert in abnormal locations on the globe [1-4]. Anomalous orbital structures may be found in cases of globe retraction not associated with Duane retraction syndrome, very severe vertical strabismus, or an elevated deficit deepening in the abduction [1,2].

In the study of Khitri and Demer, unilateral or bilateral orbital bands were found in 0.8% orthotropic and 2.4% strabismic patients [3]. It may be concluded that horizontal bands linking the MR with LR immediately posterior to the globe may limit supraduction by collision with the optic nerve and, which is particularly important, those bands are usually located too deep to be approached via conventional strabismus surgical approaches [3,4]. We must take them into account in patients with atypical restrictive strabismus. Computed tomography (CT) and, particularly, orbital magnetic resonance imaging (MRI) are essential for correct diagnosis and treatment, as the only other way to diagnose this condition is through casual findings during surgery to correct strabismus [4-6]. Regarding the axial myopia of this patient, Yamaguchi and colleagues present a series of highly myopic patients in whom magnetic resonance imaging demonstrated superotemporal globe shift out of normal relationship to the array of the rectus extraocular muscles [7]. Thus, both the superior rectus becomes abnormally nasal and the lateral rectus muscle inferior relative to the globe. Since the oculorotatory effect of a muscle is determined by pulling direction relative to the globe, superotemporal globe dislocation adds adducting and infraducting effect, while reducing supraducting and abducting effect. The muscles in the myopic patients described had no intrinsic abnormality of force generally, but muscle paths were abnormal. The severity of this anatomic abnormality is associated with the severity of strabismus and moreover, that surgical union between the corresponding margins of the superior and lateral rectus muscles can restore the normal anatomic relationship of muscles, and markedly improve the esotropia and hypotropia [7].

Some of the Questions that Arise with This Patient Are

1. Would the coexistence of an anomalous fibrous band and axial myopia fully explain the restrictive condition of the patient's ocular motility limitation?
2. If the anomalous fibrous band was congenital and generated the fixed position of ET and Hypotropia, could the axial myopia be secondary to sensory deprivation of the RE?
3. Limitation to elevation and ptosis persisted even after the restrictive factor had been eliminated and myopexia had been performed. Could there be some denervation factor added to the elevator complex as monocular deficit of elevation?

Conclusion

In conclusion, the findings in this patient with unusual restrictive strabismus could be explained by the association of axial myopia with rotated muscle axes and the existence of an abnormal band between MR and IR. The lack of improvement in elevation after surgery could be due to an added innervational factor. Imaging in restrictive strabismus helps identify associated causal factors that will be confirmed during surgery.

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