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Bilateral Type 1 Duane`S Syndrome: A Case Report

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Abstract:
Duane retraction syndrome is a strabismus syndrome characterised by congenital non-progressive horizontal ophthalmoplegia mostly affecting the abducens nerve. It mostly occurs as an isolated condition but sometimes may be associated with multiple anomalies. Here, we report a case of bilateral type 1 Duane's syndrome and its management. The patient underwent strabismus surgery in both eyes for the upshoot and alternate esotropia. She was also investigated for other possible associated anomalies. She was followed up for 6 months post-operative. The patient had a marked decrease in upshoot with orthophoria at primary position post-operatively.
Keywords: *Duane's retraction syndrome, scoliosis lumbosacral spine, mental retardation.*
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I. Introduction:

Duane syndrome is a miswiring of the extraocular muscles where the sixth cranial nerve supplying the lateral rectus muscle does not develop properly.[1] Incidence of Duane's syndrome is 1% to 4% of all strabismus cases. It presents as an isolated unilateral condition in 70% cases. In about 15% to 20% cases it may be bilateral often associated with anomalies of the skeleton, ears, eyes, kidneys, nervous system and heart. It is more common in females (60%) than males (40%).[2,3.] Here, a patient with bilateral type 1 Duane's syndrome associated with scolosis of the lumbosacral spine and mental retardation is reported.

II. Case Report:

A 15 year old female presented with complaint of decreased vision in both eyes since a couple of years and deviation of eyes since birth. She was born at full term via normal delivery at a nursing home with no significant perinatal and postnatal history. A comprehensive ocular examination revealed a visual acuity of 6/12p in the right eye improving to 6/9 with - 1.25 D cylinder @180 º and 6/9 in the left eye improving to 6/6 with - 0.50D cylinder @ 180º. A worth four dot test revealed alternate suppression and she had gross stereovision for near of 300º of an arc. She had an alternate esotropia of around 35 PD with 10 PD right hypertropia and bilateral narrowing of palpebral fissure with upshoots on adduction. Abduction was limited with widening of the palpebral fissure in both eyes (**figure 1)**. Rest of the ocular examination was normal. Cardiovascular, neurological and hearing evaluation was normal. Ultrasound abdomen and Kidney function test were within normal limits. X-ray lumbosacral spine revealed scoliosis of the lumbosacral spine.

A FDT (force duction test), FGT (force generation test) and exaggerated FGT was done under local anaesthesia. FDT was positive for medial rectus (MR), more in the right eye. FGT and exaggerated FGT were normal. The patient was operated under general anaesthesia. FDT was done intraoperatively and a very tight MR was found in both eyes. A bilateral MR recession of 5 mm and bilateral IO (inferior oblique) anteriorisation was done.

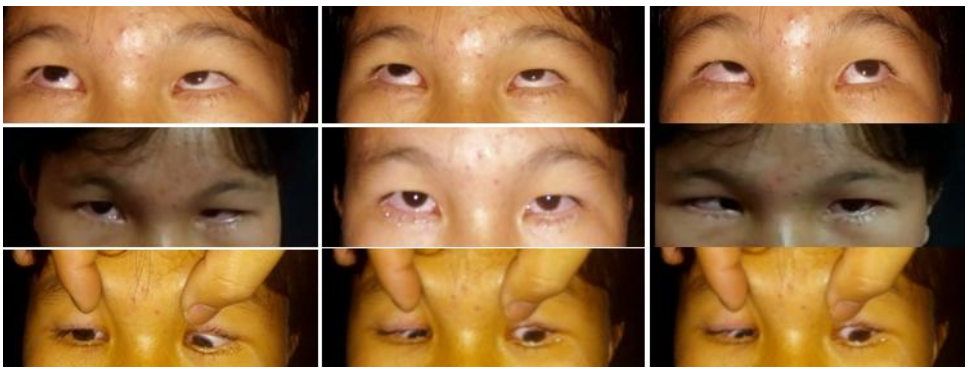

Figure 1: Pre-operative photo shows right esotropia with hypertropia, bilateral limitation of abduction and narrowing of palpebral fissure with upshoots noted in both eyes.

At 6 months follow-up, there was no horizontal deviation in primary position with a marked decrease in upshoots on adduction in both eyes. The stereovision was the same as before. **Figure 2**.

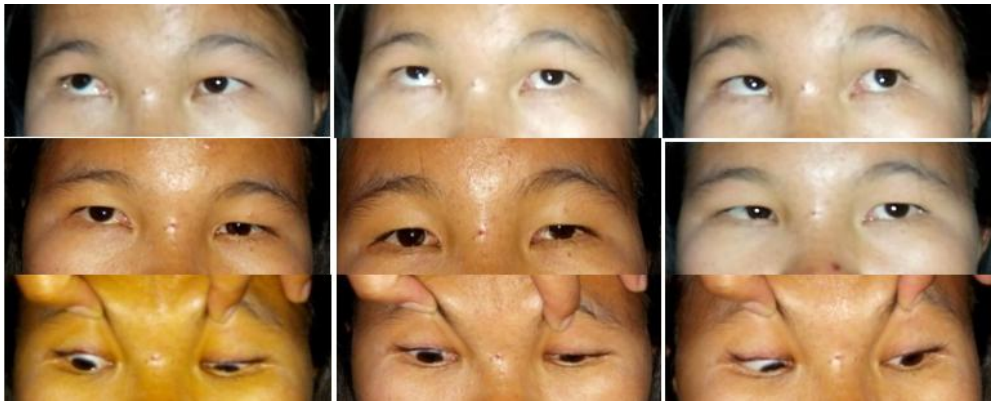

Figure 2: Post-operative photo showing orthotropia in primary gaze, bilateral limitation of abduction and a marked decrease in upshoot on adduction and levoelevation (right eye) and dextroelevation (left eye).

III. Discussion:

Duane´s retraction syndrome is a congenital and non-progressive occulomotor anomaly due to abnormal development of the 6th cranial nerve. It is characterised by limited abduction or adduction or both with retraction and narrowing of the palpebral fissure with upshoots or downshoots on adduction. There are three types of Duane`s syndrome – Type 1 (Esotropia with limited abduction); Type 2 (Exotropia with limited adduction); Type 3 (Limited adduction and abduction with Esotropia or Exotropia). Most common is type 1 which is mostly unilateral.

About 15% of Duane`s syndrome are bilateral mostly inherited as an autosomal dominant pattern.[3] The actual incidence of Type 1 bilateral Duane`s syndrome is not known as very few cases have been reported so far. Most of these are associated with other ocular anomalies (zonular cataract, aniridia, ptosis, optic nerve coloboma, crocodile tears etc.) and multiple skeletal, genitourinary, vertebral, cardiac and ear anomalies.[4,5] Here, the patient had bilateral type 1 Duane`s syndrome associated with scoliosis of the lumbosacral spine and a borderline intelligence.

Management includes a proper refractive correction, prisms glasses to improve head posture, amblyopia therapy, botox in few cases and strabismus surgery.[1,6] Surgical management is indicated when there is an abnormal head position ≥ 15 PD; Significant deviation in primary position; severe induced ptosis due to narrowing of palpebral fissure on adduction or significant upshoots or downshoots. It is important to counsel the patient that surgery does not improve motility or stereopsis.

Various approach have been used for Duane`s syndrome type 1 including medial rectus recession, posterior fixation suture on the normal medial rectus, lateral rectus resection.[7] Augmented vertical rectus transposition to the lateral rectus may improve abduction in the involved eye.[8] Recession of both horizontal recti with Y- splitting of lateral rectus has been an effective procedure for correcting the horizontal deviation with upshoots and downshoots.[9] Inferior oblique (IO) anteriorization is reported to be effective in treating upshoots. Here, the IO is transposed just temporal and posterior to the inferior rectus insertion to avoid the anti-elevation syndrome. However, IO anteriorization may worsen the downshoots.[10]

IV. Conclusion:

Duane`s syndrome is a rare form of strabismus entity with bilateral type much rarer. A complete ocular and systemic evaluation should be done to rule out associated anomalies. Conservative management can be done in many cases. Surgical management should be done only when required taking into account the limitations of the surgery with an informed consent of the patient.

The patient underwent bilateral medial rectus recession and IO anteriorization under general anaesthesia. At 6 months follow-up, the patient had no horizontal deviation with a marked reduction in upshoots. Hence, IO anteriorization is an effective method for managing upshoots and can be an alternative of lateral rectus recession with Y-splitting. It may be combined with medial rectus recession for the horizontal deviation in type 1 Duane`s as done in this case. The long-term result is yet to be seen with further follow-up of the patient. A study with larger number of patients with longer follow-up would be ideal but due to the rare occurrence of the condition, it may take a long time to come up with a definite conclusion.

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