

## Original Article

## MANAGEMENT OF DUANE RETRACTION SYNDROME

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**Abstract**

**Purpose:** To determine the clinical profile of various subtypes of Duane retraction syndrome patients and to evaluate different surgical strategies in correction of different clinical signs.

**Patients and Methods:** This was a hospital-based prospective interventional study conducted at the Ophthalmology department, Al-Azhar university hospital in Assiut over a duration of two years from October 2017 to October 2019. Thirty one patients of DRS were scheduled to be included in this study. They were divided into three groups; Group I: 21 patients of DRS type I, Group II: 4 patients of DRS type II and Group III: 6 patients of DRS type III. Age of patients ranged from 9 months to 28 yrs old. Postoperative results were evaluated over 6<sup>th</sup> months follow-up period. **Results:** Thirteen patients were left without surgical intervention, they were orthophoric or with minimal misalignment in PP with mild retraction of the globe on attempted adduction and none of them had any vertical shoot nor abnormal head position, eighteen patients of DRS had surgical intervention. Nine cases were exotropic, 8 cases were esotropic & 1 case was orthophoric. Horizontal deviations were managed with unilateral or bilateral muscle recession & simultaneous recession of MR & LR. Retraction of the globe was managed with single muscle recession or simultaneous recession of both MR & LR. Vertical shoots were managed with LR Y-splitting, IO recession & simultaneous recession of both MR & LR. **Conclusion:** Every patient of Duane retraction syndrome has an individual story, with good diagnosis & accurate grading to different signs we can determine the best surgical technique for the correctable signs. Single muscle recession & simultaneous recession of both MR & LR can correct horizontal strabismus in PP, the retraction of the globe on attempted adduction & the abnormal head position effectively. Overshoots can be corrected by LR Y-splitting, simultaneous recession of both MR & LR IO anteriorisation surgery is the best choice in cases with HT in PP & slow upshoot. Small LR resection can be done in esotropic cases with mild retraction (grade 1).

**Keywords:** Duane Retraction Syndrome, Strabismus surgery, Esotropia, Exotropia

**1. Introduction**

Duane Retraction Syndrome (DRS) is an abnormal pattern of ocular motility characterized by retraction of the globe with narrowing of the palpebral fissure on attempted adduction and a variety of other abnormal movement of the affected eye when

the other eye fixates in various cardinal positions [1,2]. The abnormal pattern of ocular motility of DRS is the result of developmental adaptation that occurs in the embryo as consequence of the Electromyography studies have shown paradoxical

innervations of lateral rectus muscle and anomalous synergistic innervations of medial rectus, inferior rectus, superior rectus

### 1.1. Huber classification

This classification [1,4] includes three types; they are (**type 1**); esotropia in PP, restrict abduction (Most common type).

### 1.2. Clinical manifestations

The clinical manifestations in DRS can be quite variable and will depend upon the wide array of innervational abnormalities of the lateral rectus. The most important clinical features recognized in these patients include the following:

*Strabismus:* Although many patients with DRS can orthophoric in primary position, many patients are esotropic and a few can be exotropic.

*Eyelid fissure changes and globe retraction:* Narrowing of the palpebral fissure in adduction occurs as a result of passive movement of the eyelids over the retracting globe but is also due to a decrease in electrical activity seen in the levator muscle on adduction as demonstrated by simultaneous electromyographic recordings of the levator and medial rectus muscles.

### 1.3. Management

Non surgical approach is will normalize rotations. The main goals of surgery are to centralize and expand the field of single binocular vision. *For patients with type 1 Duane retraction syndrome:* Ipsilateral medial rectus recession or bilateral medial rectus recession for deviations larger than 20 prism diopters. Most surgeons don't favor resection of lateral rectus because of the likelihood of that globe retraction will worsen. Partial or full lateral transposition of both of the vertical rectus muscles or the superior rectus alone with or without posterior sclera fixation (myopexy) has been found to improve abduction. *For patients with type 2 Duane syndrome:* Unilateral or

and oblique muscles [3,4,5] as a result of absence or hypoplasia of 6<sup>th</sup> cranial nerve [6,7].

(**type 2**), poor adduction and exotropia and (**type 3**); limited adduction and abduction.

*Compensatory head posture. Upshoots and downshoots:* Upshoots and downshoots are sudden vertical deviations that occur in DRS patients when the eye begins to move up or down in the adducted position. *Alphabetical patterns:* A large incidence of increased innervation of the lateral rectus in elevation or depression explains the frequent observation of A, V, or X patterns in these patients.

*Sensory anomalies:* Most patients with DRS adopt a compensatory head turn when the eyes are not aligned in primary position to maintain single binocular vision. Even if suppression can be seen in some patients, most of them are aware of a second image when questioned, but they usually tend to ignore this second image [8].

bilateral lateral rectus recession for large-angle exotropia. *Patients with type 3 Duane syndrome:* Often have straight eyes in or near the primary position and little, if any head turn. Severe globe retraction may be lessened by recession of both the medial and lateral rectus; this is also an option in treating retraction in type 1 and type 2 Duane retraction syndrome. Procedures to address the upshoot or downshoot include: large lateral rectus recession with splitting in Y configuration, retro-equatorial fixation of the lateral rectus muscle, and more recently, disinsertion of the lateral rectus muscle and reattachment to the lateral periosteum of the orbit [9].

## 2. Patients and Methods

This was a hospital-based prospective interventional study conducted at the Ophthalmology dept., Al-Azhar Univ. hospital in Assiut over duration of two years from October 2017 to October 2019. Thirty

one patients of DRS were scheduled to be included in this study. *Inclusion criteria:* patients with Duane retraction syndrome. *Exclusion criteria:* patients with history of previous ocular muscle surgery.

## 2.1. Preoperative evaluation

*Detailed ocular examination and diagnosis*; taking history: time & onset of the disease, previous photograph, developmental milestones, past history of ocular trauma, glasses, occlusion therapy strabismus surgery & anasthaetic complications. Visual acuity testing in preverbal children using Occlusion of one eye (fixation preference), Fixation toy: C.S.M (central, steady, maintained) and Fixation test (cover/uncover test for manifest deviation or vertical prism deviation test if no deviation). But in preliterate children we used Tumpling E chart and Lea symbol chart and in literate patients we used Landolt's chart. Ocular motility examination of versions and ductions in 9 gazes using pen light torch using (Hirschberg test (corneal reflex test), Cover uncover test, Alternating cover test and prism alternating cover test in near and far distances using single plastic prisms. Lid examination: look for abnormalities, Anterior segment examination with the help of standard slit lamp, Cycloplegic refraction using eye drops containing cyclopentolate

## 2.2. Operative procedures

Informed consent from the patients or parents was taken before surgery with permission of taking preoperative and post-operative photo pictures. All the patients were operated under general anasthaesia by the same surgeon (Hadeel Raif) types of surgical techniques: Unilateral or bilateral MR recession, Unilateral or bilateral LR recession, Simultaneous recession of both MR & LR, LR recession with split, small LR resection and I O recession +4 (at the same level of IR insertion). General surgical principles; \* Proper positioning of the patient so that the head is in extended position (the chin is higher than the forehead) \* Guard against infection using povidone iodine in sterilization. \* Limbal conjunctival

### 2.2.1. LR Y-splitting technique

By dissection of the LR muscle into 2 halves by the hook then separating it 12 mm apart then suturing the recessed

& phenylephrine as active ingredients and posterior segment examination for (Retina, optic nerve) using indirect ophthalmoscope. Subtyping of Duane retraction syndrome cases according to Hubers classification. Thirty one patients were divided into 3 sub-groups:

**Group I:** 21 patients with DRS type I.

**Group II:** 4 patients with DRS type II.

**Group III:** 6 patients of DRS type III.

Non-surgical management included glasses prescription to 3 cases (2 hypermetropic and 1 myopic). Selection of cases that required surgical intervention was made according to presence of one or more of the following: \* Strabismus in 1ry position not corrected by glasses. \* Abnormal head position. \* Retraction of the globe on adduction (grade 2 or more). \* Overshoot of the globe on attempted adduction (grade 2 or more). Surgical techniques were guided by Kenneth Wright number adjustment for common strabismus [10]. Proper physical fitness was done to all cases.

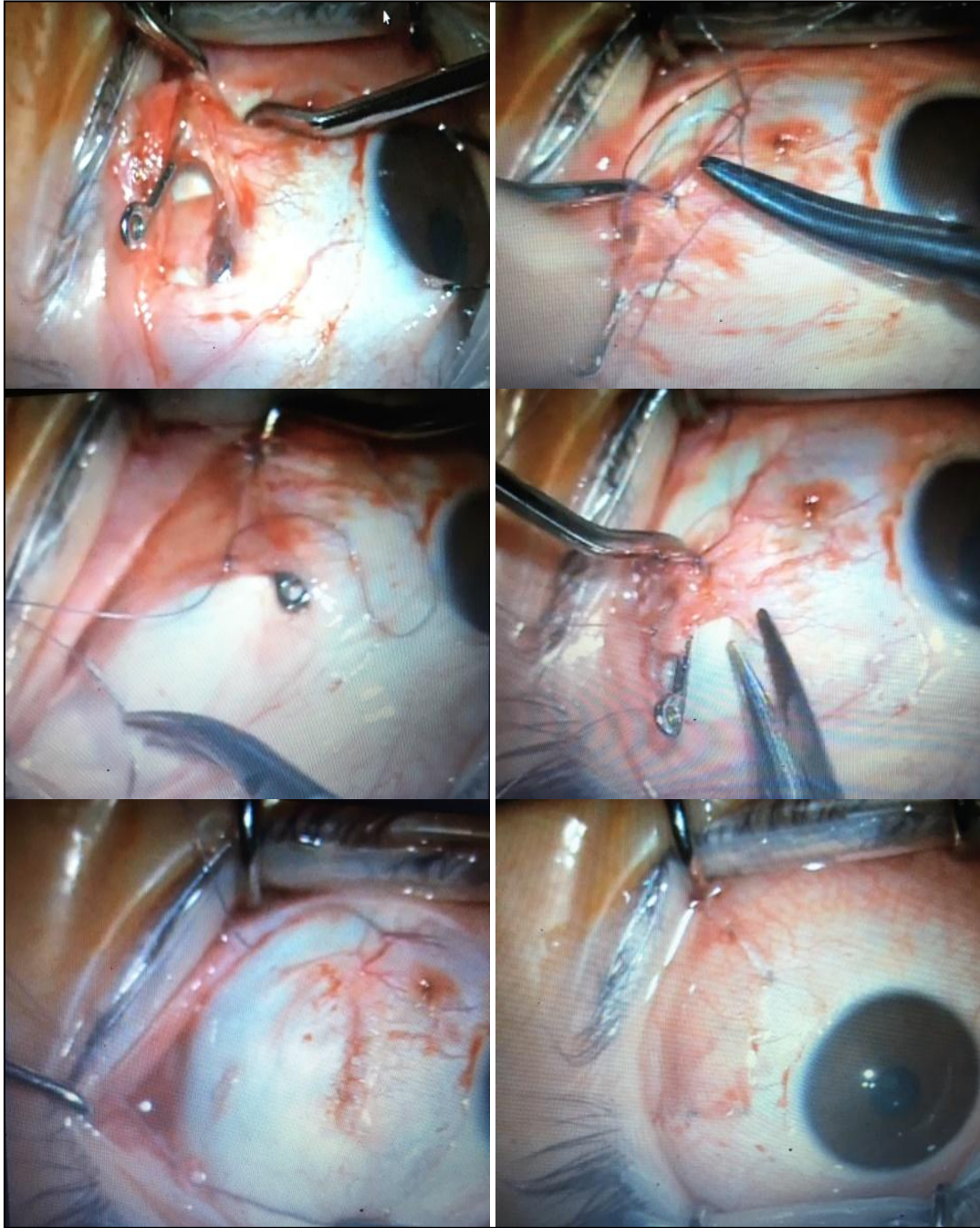
incision 6 mm with 2 relaxing incisions.

\* Hooking the muscle by stevens hook.

\* Avoid penetration of posterior tenon capsule & protrusion of orbital fat. \* Muscle suturing by double armed absorbable suture after removal of anterior tenon capsule.

\* The muscle fibers is well secured by one partial thickness in the superficial fibers and the other is full thickness with good lock bite on each pole. \* Scleral sutures behind the new insertion using spatulated needle. \* Avoid superficial scleral suture (easily cut & may lead to slipped muscle) and avoid very deep one (can lead to perforation & retinal detachment). \* Good muscle dissection by removing the intramuscular septum & check ligaments.

halves behind the new insertion in the sclera, fig. (1).



**Figure 1.** LR Y-splitting surgery

### 2.2.2. IO recession technique

Moving the IO muscle insertion at the same level of the IR muscle insertion

### 2.3. Follow up

Prescription of routine postoperative treatment: systemic antibiotic & anti-inflammatory for 1 week and combination of topical steroid & antibiotic for 4 weeks.

#### 2.3.1. DRS type I

DRS type I includes 21 patients; three cases were presented as examples.

(IO graded recession +4). So the IO muscle will be depressor not elevator.

Follow up of all patients on 1<sup>st</sup> post-operative day, after one week, 2 weeks & 4 weeks then every month till 6 months.

They are case-1, fig. (2), case-2, fig. (3) & case-3, fig. (4)

**a.** (preoperative)



**b.** (postoperative)



**Figure 2.** Orthophoric DRS type I; **a.** f 10 yr old, Lt DRS type I, orthophoric in PP, Lt limited abduction with retraction of the globe grade 4 on attempted adduction. Decision: simultaneous C of both MR 5mm & LR 7 mm in Lt eye, **b.** postoperative result: Orthophoric in PP, improvement of retraction to grade 1 .

**a.** (preoperative)



**b.** (postoperative)



**Figure 3:** Esotropic DRS type I **a.** f 2 yrs old, Rt DRS type I , ET 35 PD in PP, limited abduction & retraction of the globe grade 2 in Lt eye on attempted adduction. Decision: Rt MR C 6 mm. & LR S 3.5 mm, **b.** (postoperative results): orthophoric in PP, Improvement of retraction to grade 1.



**Figure 4.** Orthophoric DRS type I. F 11 yrs old with Lt DRS type I, orthophoria in PP, limited abduction grade (-3) & Retraction of the globe grade (1) on attempted adduction in Lt eye, Decision: follow up , no surgical intervention.

### 2.3.2. DRS type II

DRS type 2 includes 4 patients; three cases were presented as examples.

They are case-1, fig. (5), case-2, fig. (6) & case-3, fig. (7).

#### a. (preoperative)



#### b. (postoperative)



**Figure 5.** Exotropic DRS type II; **a.** f 2 yrs old, Lt DRS type II , Lt XT 20 PD in PP , Lt limited adduction, retraction of the globe grade 2 & upshoot grade 4 on attempted adduction & face turn to the Rt shoulder. Decision: simultaneous C of both MR 4 mm & LR 8 mm with split 16 mm in Lt eye, **b.** postoperative result: Orthophoria in PP, Improvement of retraction of the globe to grade 0 & upshoot to grade 0 with elimination of face turn.



**Figure 6.** Orthophoric DRS type II M 13 yrs old with bilateral DRS type II, XT 10 PD in PP, limited adduction & retraction of the globe grade 1 on attempted adduction in both eyes. Decision: follow up, no surgical intervention.



**Figure 7.** Patient No.4, group II F 3 yrs old with Lt DRS type II, orthophoria in PP, limited adduction & Retraction of the globe grade (1) on attempted adduction in Lt eye, Decision: follow up, no surgical intervention.

### 2.3.3. DRS type III

DRS type 3 includes 6 patients; one case one case of Esotropic DRS only

was presented as an example. It is case-1, fig. (8).

a. (preoperative)



b. (postoperative)



**Figure 8.** Esotropic DRS type III; **a.** M 1yr old, Lt DRS type III, LT ET 20 PD in PP, Lt limited adduction with retraction grade 3 & upshoot grade 3 of the globe with attempted adduction. Decision: simultaneous recession of both MR 6 mm & LR 5 mm in Lt eye, **b.** postoperative result: Orthophoric in PP, improvement of retraction to grade 1 and up-shoot of the globe to grade 1 with attempted adduction

## 2. Results

A total of 31 patients with DRS, 18 females (58%) & 13 Males (42%). 65% of them had the disease in Lt eye, 16 % in Rt eye and 19 % bilateral. 69% presented with type I DRS, 12% presented with type II Duane retraction syndrome & 19% of cases presented with type III DRS, tab<sub>s</sub>. (1, 2, 3 & 4). Strabismus in PP. represented as 8 esotropic cases (26 %), 9 exotropic cases (29%) and 16 orthophoric cases (45%). the mean preoperative angle of ET in PP. was 33.2 PD & postoperative angle was -1.3 PD, (P-value <0.001), tab. (5). The mean preoperative angle of XT in PP. was -24.4 PD and the mean postoperative angle was 0.89 PD, (P-value < 0.001). HT was found in 2 patients (6.5%),

one of them esotropic and the other was exotropic, tab. (6, 7 & 8). The retraction of the globe was found in 22 eyes (62.8%), the preoperative grading mean  $2.5 \pm 0.8$  & the postoperative grading mean was  $0.9 \pm 0.61$  (P-value <0.001). Overshoot of the globe with attempted adduction was found in 11 cases (31%) (10 cases upshoot & 1 case downshoot), the preoperative mean of upshoot cases was  $2.8 \pm 1$  & the postoperative mean  $0.8 \pm 0.63$  (P-value <0.001) (9, 10, 11 & 12). Abnormal head posture was found in 7 cases (22.5%), 6 to the opposite side & 1 to the same side, all of them improved completely. 3 cases had glasses prescription (9.6%), 18 patients (58%) had surgical intervention, tab<sub>s</sub>. (13).

**Table 1.** Gender distribution

Gender	No. of cases
Male	13
Female	18

**Table 2.** Age distribution

Minimum	0.75 yrs
Maximum	28 yrs
Range	27.25

**Table 3.** Laterality

Laterality	No. of cases
Rt	5
Lt	20
Bilateral	6

**Table 4:** Classification

Classification	No. of Cases
Type I	21
Type II	4
Type III	6

**Table 5:** Strabismus in PP.

Strabismus in PP	No. of cases
Cases with orthophoria or minimal misalignment less than 10 PD	14
Esotropic DRS	8
Exotropic DRS	9

**Table 6:** postoperative results of strabismus in PP

Ocular deviation in 1ry position	Post-operative result
ET in 7 cases	Orthophoria in 5 cases XT 10 PD in 2 cases
ET with HT in 1 case	Orthophoria
XT in 8 cases	Orthophoria in 8 cases
XT with HT in 1 case	Orthophoria

**Table 7:** Esotropic DRS

	Minimum	maximum	range	Mean	SD	P-value
Preoperative ET	ET 20 PD	ET 55 PD	ET 35 PD	ET 33.8 PD	15	0.0005
Postoperative result	0 PD	XT -10 PD	10 PD	-1.3PD	3.5	

**Table 8:** Exotropic DRS

	Minimum	maximum	Range	Mean	SD	P-value
Preoperative XT	XT -20 PD	XT -30 PD	XT -10 PD	XT -24.4 PD	4.6	0.0005
Postoperative result	0 PD	ET 8 PD	8 PD	0.89 PD	2.7	

**Table 9:** Retraction of the globe results

Eye No.	Retraction grade	Postoperative grade	Eye No.	Retraction grade	Postoperative grade
1	1	1	12	2	0
2	2	1	13	3	3
3	2	1	14	3	1
4	2	1	15	3	1
5	2	1	16	3	1
6	2	1	17	3	1
7	2	1	18	3	1
8	2	1	19	3	1
9	2	1	20	4	1
10	2	0	21	4	1
11	2	0	22	4	0

The retraction of the globe was found in 22 eyes (62.8%)

**Table 10:** Retraction of the globe results

Preoperative grade (Mean ±SD)	Postoperative grade (Mean ±SD)	p-value
2.5±0.8	0.9± 0.61	0.0005



**Table 11: overshoots results**

Eye No.	Up-shoot preoperative grade	Postoperative grade
1	1	1
2	2	1
3	2	1
4	2	1
5	3	0
6	3	1
7	3	2
8	4	0
9	4	0
10	4	1
Eye No.	Down-shoot grade	Postoperative Grade
1	3	1

Upshoot of the globe on attempted adduction was found in 10 cases (32.2%)

**Table 12: overshoots results**

Up-shoot preoperative grade (Mean ±SD)	Postoperative grade (Mean ±SD)	p-value
2.8± 1	0.8 ± 0.63	<b>0.002</b>

**Table 13: Abnormal head position**

Face turn	Post-operative result
Face turn to the opposite side in 6 exotropic cases	Improved completely
Face turn to the same side in one esotropic case	Improved completely

### 3. Discussion

#### 3.1. Principles of surgical approach in DRS

##### 3.1.1. Esotropic DRS

Kekunnaya et al [11] and Doyle & Hunter [12] Consider recession of the medial rectus muscle or lateral transposition of one or both vertical rectus muscles (with or without simultaneous weakening of the medial rectus muscle by recession or botulinum toxin injections). Vertical rectus muscle transposition may be augmented by simultaneous resection of the transposed muscles or by placing posterior augmentation sutures on the transposed muscles. When globe retraction is mild, recession of the medial rectus muscle may be combined with a modest resection of the lateral rectus muscle. If globe retraction is severe and creates a deformity, consider recession of both the medial and lateral rectus muscles. Contralateral medial rectus recession may be added for large deviations [11,12]. Cases with bilateral esotropic DRS, which is the commonest presentation [13], are most often corrected by bilateral MR recessions, depending on

the primary position deviation, muscle tightness and limitation of ductions. Sachdeva et al. [14] studied 14 patients with bilateral esotropic DRS and found 86% success rate with a mean surgical dose of 5.6 mm (range being 5-7 mm) for bi-MR recessions. The mean preoperative esotropia in this series was 38 PD, with none of the patients developing significant postoperative adduction limitation. Another technique to correct the eso-deviation and improve abduction in these eyes is by transposition of vertical rectus muscles to the LR along the spiral of Tillaux. This may or may not be combined with MR recession, as there is risk of inducing anterior segment ischemia. There is also a risk of inducing new vertical or torsional deviations in these eyes, the most common being a hypotropia [15]. To prevent this, intraoperative FDT meticulous dissection of the vertical rectus to release them from the lid retractors and intraoperative monitoring

of torsion is recommended after FDT. In 2006, Johnston et al [16] described the role of superior rectus transposition (SRT) to the LR with or without MR recession. In their analyses of 52 patients with type I esotropic DRS, the preoperative deviation (range: 10-30 PD) improved to within 10 PD of esotropia in 95% of patients. Head turn improved in all cases, with

**3.1.2. Exotropic DRS:**  
Kekunnaya et al [11] and Doyle & Hunter [12] Consider recession of the ipsilateral lateral rectus muscle in most cases. In more severe cases, a large lateral rectus recession may be combined with transposition of one or both vertical rectus muscles [10,11]. Theodorou and Burke [17] treated 11 patients with bilateral

### **3.2. Globe retraction**

Theodorou and Burke [17] considered recessions of the co-contracting muscles to correct globe retraction. MR recession ranging from 5 to 6.5 mm and LR ranging from 7 to 9 mm or sometimes even periosteal fixation may be required. When primary position esotropia is present, MR should be recessed more than LR; if

### **3.3. Overshoots**

Upshoots and downshoots in DRS may be due to the mechanical effect caused by tight muscles or due to innervational anomalies. Kekunnaya et al [11] and Doyle & Hunter [12] consider Y-splitting of the lateral rectus muscle reduces upshoot and downshoot in adduction without altering the alignment in primary gaze. Rogers and Bremner [19] have described Y splitting of the LR with or without its recession to treat overshoots caused by this mechanical leash effect. With Y split, each muscle half balances the other; thus, when the eye elevates in adduction, the lower arm contracts and prevents the globe from suddenly slipping up and vice versa in depression. Awadein [20] studied the role of inferior oblique (IO) weakening in all types of DRS which presented with a slow upshoot in which the movement was similar to IO overaction. He found that IO myectomy with or without horizontal rectus recession

complete elimination in 27% of the patients. Preoperative abduction limitation was noted to be from -3 to -4 in all eyes, which increased by 15°-45° after SRT. They also noted that there were no induced vertical deviations after SRT, unlike those seen after transposing both the vertical rectus muscles.

exotropic DRS. Unilateral LR recession (3-18 mm recessions for deviations ranging from 12 to 60 PD) with conjunctival recession was the commonest surgery done, and cases with significant globe retraction also underwent additional small MR recession.

not, then LR can be recessed 1 mm more than MR. It has been suggested that adults may need larger recessions than pediatric patients due to long-standing globe retraction causing orbital tissue changes, and retraction may recur due to continued co-contraction of the muscles [18].

improved upshoot in this set of patients without inducing any vertical deviations or IO underaction. So we are agreeing with Kekunnaya et al [11] and Doyle & Hunter [12] in management of strabismus in PP, abnormal head position and retraction of the globe by single muscle recession or bilateral recession of MR or LR. We are agreeing also with Theodorou N, Burke J [17] in correction of globe retraction by simultaneous recession of both co-contracting muscles but we recessed LR more than MR by 2 mm in orthophoric cases, LR more than MR by 4 mm in presence of XT 20 PD. We are agreeing with Kekunnaya et al [11] and Doyle & Hunter [12] in correction of overshoots by LR Y-splitting and we are agreeing with Awadein [20] in correcting slow upshoot by IO weakening but we considered IO anteriorisation as a best choice to correct slow upshoot with hypotropia in PP.

#### 4. Conclusion

Every patient Duane retraction syndrome has an individual story, with good diagnosis & accurate grading to different signs we can determine the best surgical technique for the correctable signs. Single muscle recession & simultaneous recession of both MR & LR can correct horizontal strabismus in PP, the retraction of the globe on attempted adduction & the abnormal head position effectively. Overshoots can be corrected by LR Y-splitting, simultaneous recession of both MR& LR. IO recession surgery is the best choice in cases with HT in PP & slow upshoot. Small LR resection can be done in esotropic cases with mild retraction (grade 1).

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