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Comparative Study of Recession with Y- splitting Vs Recession Only of Lateral Rectus Muscle Surgery for up/down Shoot in Duane Retraction Syndrome

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Authors' contributions

This work was done in collaboration between both authors. Author VCR designed the study, wrote the protocol, performed statistical analysis and wrote the daft of the manuscript. Author SCR corrected the draft, formatted the paper in the template of the journal and contributed in the discussion. Both authors read and approved the final manuscript.

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ABSTRACT

Purpose: To evaluate and compare the effectiveness of lateral rectus muscle recession with Y-splitting versus recession only in the surgical treatment of up/down shoot in Duane retraction syndrome patients.
Material and Methods: In this prospective study, 42 patients of Duane retraction syndrome with up/down shoot underwent surgery over a period of seven years, were divided into two groups of twenty one each. In group A, Y-splitting of lateral rectus with recession and in group B, only lateral rectus recession was performed. In all the patients width of lateral rectus muscle at its insertion was measured. Postoperative results were compared and analyzed in terms of up/down shoot on adduction. Data analysis was performed using R Core Team (2020) R., and the P value <0.05 was taken as significant.
Results: The Mean age at the time of surgery was 12.5 ± 4.5 years in group A, and 11.5 ± 3.9

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years in group B (range, 5 to 22 years). Correction of up/down shoot was 100% in group A and 81% in group B. Five patients in group B showed no improvement of up/down shoot in whom the width of lateral rectus was 7mm or less and the surgery was recession of lateral rectus only. In these 5 patients, Y-splitting of lateral rectus was done in second sitting with good result Mean follow up was 12 months (range, 3 weeks to 3 years).

Conclusions: Recession of lateral rectus muscle with Y-splitting is more effective (100%) when compared to recession only, in correcting significant up/down shoot in Duane retraction syndrome. Lateral rectus muscle Y- splitting is not necessary in these patients with upshoot / down shoot if the width of the muscle at insertion is within normal limits (8.3 -12.5 mm). In these cases simple recession is enough. Lateral rectus muscle Y splitting is essential when the width of lateral rectus muscle at insertion is 7mm or less.

Keywords: Duane retraction syndrome; lateral rectus recession; lateral rectus Y- splitting surgery.

1. INTRODUCTION

Duane retraction syndrome (DRS), also known as Stilling-Turk-Duane syndrome, is defined as congenital miswiring of lateral and medial recti muscles, resulting in an impaired ocular motility syndrome that includes palpebral fissure narrowing. DRS has been classified among the congenital cranial dysinnervation disorders (CCDD). Its incidence is approximately 1-4%of the total cases of strabismus [1,2]. The incidence of upshoot & down shoot in DRS population is about 43% [3]. DRS is characterized by (i) complete or partial absence of abduction of the affected eye, (ii) partial or rarely complete deficiency of adduction of the affected eye, (iii) retraction of the affected eye into the orbit on attempted adduction with narrowing of palpebral fissure, and (iv) in some patients on attempted adduction, upshoot or down shoot of the affected eye [4,5,6].

The up and down shoot is believed to be caused by “bridle effect” [6] of tight lateral rectus (LR) or “leash effect” [7] or aberrant innervation of vertical muscles [8]. When eye adducts and moves above or below the horizontal plane, there is sudden slippage of the tight LR, causing an upshoot or down shoot [9]. In severe cases, this has been described to manifest even with the slightest of movement in adduction, known as “knife edge effect” [7]. Disfigurement due to up/down shoot on attempted adduction is not acceptable both cosmetically and functionally and is an absolute indication for correction. In recent times most commonly used surgical methods for the management of upshoot or down shoot in DRS are (i) simple recession of lateral rectus [10] and (ii) Y- splitting of lateral rectus with recession [11,12,13].

Only three studies comparing the Y-splitting and recession versus recession of lateral rectus muscle for up/down shoot of Duane retraction syndrome are available from countries outside India [14,15,16]. However, to the best of our knowledge there is no published data on this comparative study from India. The aim of this prospective study was to evaluate, compare and analyze the results of these two surgical procedures in correcting the up/down shoot in DRS.

2. SUBJECTS AND METHODS

In this prospective interventional cohort study of 42 patients of DRS with up/down shoot, with or without horizontal deviation (both genders of all age groups), presenting to the department of Squint & Pediatric ophthalmology unit of a tertiary eye institute over a period of seven years (2003-2009) were included. Sarojini Devi Eye Hospital is the teaching hospital for undergraduate and post graduate students of Osmania Medical College and Gandhi Medical College in Hyderabad. It has 500 beds and facilities for treatment of eye patients of ophthalmology subspecialties.

Patients with previous squint surgery were not included in this study. All the patients were subjected to visual acuity testing, refraction and subjective correction, detailed orthoptic evaluation pre and post operatively. The severity of upshoot on adduction were graded on a 4-point scale: 1 indicating minimal upshot, 4 indicating severe upshoot where cornea is not visible (Fig. 1 a, b, c, d).

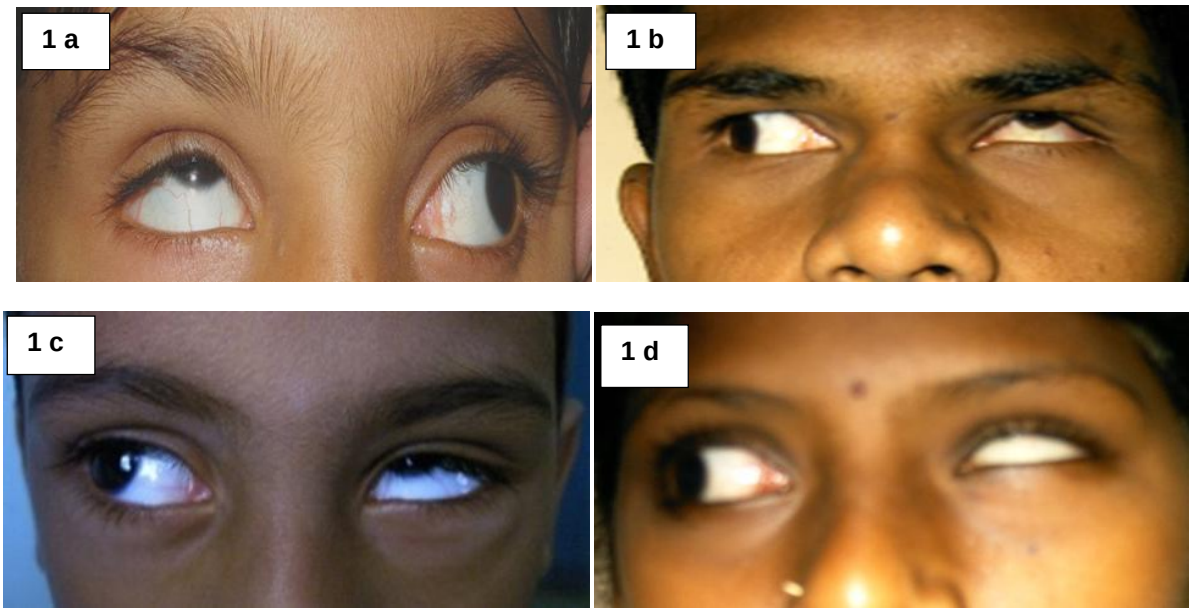

Fig. 1. Showing the grading of upshoot on adduction (a) Right eye - grade 1 upshoot, (b) Left eye - grade 2 upshoot, (c) Left eye- grade 3 upshoot, (d) Left eye - grade 4 upshoot where cornea is not visible

The patients were divided into two groups of 21 each keeping the age, sex, and laterality in appropriate ratio. In patients of Group A, Y-splitting of lateral rectus was performed along with requisite amount of recession. In Group B, only lateral rectus recession was performed. In all the patients the width of the lateral rectus muscle at its insertion were measured. The mean follow up was 12 months (range, 3 weeks to 3 years). Postoperative results were compared and analyzed.

2.1 Surgical Procedure

All the surgeries were performed under general anaesthesia and limbal approach was preferred for isolation of rectus muscles to have more surgical space. All the surgeries were done by single surgeon (VCR). Forced duction test was done in all patients. In Group A patients, the lateral rectus muscle was split horizontally into superior and inferior equal halves from its insertion, along the length of the muscle for 10mm, and the two split ends of the muscle were anchored to the sclera separately with 6-0 vicryl making the new insertion as 12 –14 mm wide with required amount of recession (Fig. 2b). Normal width of lateral rectus muscle at insertion is 8.3 to 12.5 mm [17,18]. In Group B patients, the lateral rectus muscle was recessed to the requisite amount like in the traditional standard procedure (Fig 2a).

Adequate recession of medial rectus was done in all the cases to reduce globe retraction & horizontal deviation. Amount of recession was decided depending upon the tightness of muscles observed intraoperatively, appearance of face turn, up/down shoot and palpebral fissure narrowing. In all the patients, the width of the lateral rectus muscle was measured with calipers during surgery. Post-operatively patients were given antibiotic steroid eye drops three times daily in the operated eye, serratio peptidase tablets three times daily for one week. At the time of discharge, steroid antibiotic eye drops were continued for two times daily and tapered in 3 weeks. After the operation eye was examined for up/down shoot on adduction, retraction and conjunctival opposition were observed. Patients were followed up one week, three weeks, two months and six months post operatively.

2.2 Statistical Analysis

Data analysis was performed using R Core Team (2020) R. We used t-test, Chi-square test, Ranksum test or Fisher's exact test based on the distribution and type of data, to compare pre-op and post-op parameters between the two group (R+S and REC). We used Wilcoxon signed rank test to compare pre-op and post-op up/down shoot within recession +splitting (R+S) and recession (REC).The P value of <0.05 was taken as significant.

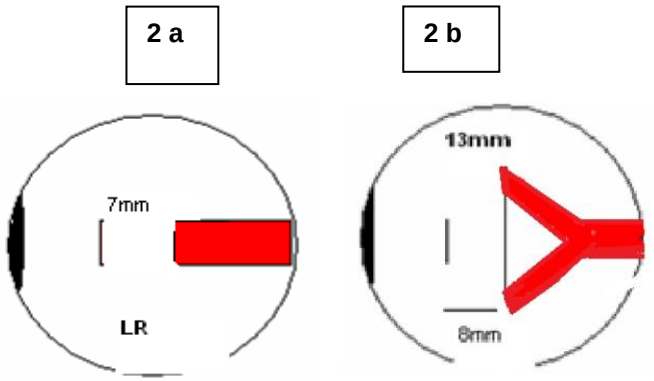

Fig. 2. Showing schematic drawing of (a) lateral rectus recession (b) lateral. Rectus Y- splitting and recession. Post split lateral rectus width increased to 13 mm from 7 mm

Table 1. Summary of ocular findings 42 patients

Patient group	M/F	Right eye	Left eye	Exo	Eso	Ortho	Up shoot	Up and down shoot #
Group A (21)	6/15	7	14	13	1	7	16	5
Group B (21)	5/16	6	15	12	0	9	16	5
Total (42)	11/31	13	29	25	1	16	32	10

M= Male, F= Female, Exo= Exotropia, Eso= Esotropia, Ortho= Orthotropia
 # the findings of upshot and down shoot in the affected eye of the same patient (Fig. 3)

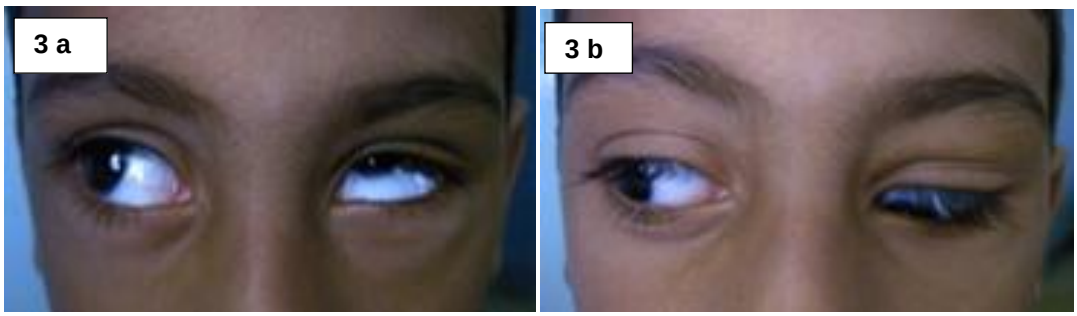

Fig. 3. Showing (a) sometime upshot of left eye on adduction and (b) other time downshoot of left eye on adduction in the same patient

3. RESULTS

Forty two patients (31 female,74% and 11 male, 26%) were operated for DRS. The mean age of the patients was 11 ± 6.9 years (range 5-22 years). DRS was present in 29 (69%) patients in left eye and the rest 13 (31%) in right eye. In primary gaze 25 (59.5%) patients had exotropia (deviation of eye away from the nose) and 1 patient had esotropia (deviation of eye towards the nose); while 16 (38%) patients had orthotropia (both eyes straight without any deviation), (Table 1).

All the 42 cases were congenital. DRS with up/down shoot was more common in females (74%) and left eye was more often involved (69%). Refractive error with mild amblyopia was seen in 7 patients (16%) and the remaining were emetropic. In 16 patients stereopsis was present with Titmus charts testing. In 21 patients recession of lateral rectus with splitting was done, remaining 21 received only recession of lateral rectus. The width of lateral rectus muscle was 7mm or less in 12 patients, and 8-12 mm in 30 patients.

Among the 12 patients in whom the width of lateral rectus was 7mm or less; 7 were in group A who underwent recession with splitting and 5 were in group B in whom only recession was done. Among 30 patients in whom lateral rectus muscle width was 8-12 mm (normal) 16 were in group A who had recession of lateral rectus with splitting while 14 belong to group B who had only recession of lateral rectus. The width of lateral rectus muscle insertion varied from 6 to 11mm. The values of different variables and their statistical tests are listed in Table 2.

When the results of the two groups were analyzed, it was found that the correction of up/down shoot was very satisfactory in all the patients (Fig 4,5), except in 5 patients of group B in whom the width of lateral rectus at insertion was 7mm or less and the surgery was only recession of lateral rectus.

Table 2. Statistical tests and their p values of different variables

Variables	Group A R+S (21)	Group B REC (21)	Statistical Test	P value
Age in years, mean (SD)	12.5 ± 4.5 years	11.5± 3.9 years	t-test (40 df)=0.77	0.445
Gender				
Female	17 (81)	14 (66.7)	Chisq. (1 df)	0.483
Male	4 (19)	7 (33.3)	= 0.49	
Eye				
Left eye	12 (57.1)	15 (71.4)	Chisq. (1 df)	0.52
Right eye	9 (42.9)	6 (28.6)	= 0.41	
PreOp. median IQR	3 (3,3)	4 (3,4)	Ranksum test	0.025
PostOp. median IQR	0 (0,0)	0 (0,0)	Ranksum test	0.02
Lateral rectus insertion width Intraoperative. median IQR	8.5 (6.5,9)	8.5 (8.5,9)	Ranksum test	0.908
OutCome				
Success	21 (100)	16 (76.2)	Fisher's	0.048
Failure	0 (0)	5 (23.8)	exact test	

R+S=Recession +splitting of lateral rectus, REC=Recession of lateral rectus, IQR=Interquartile range

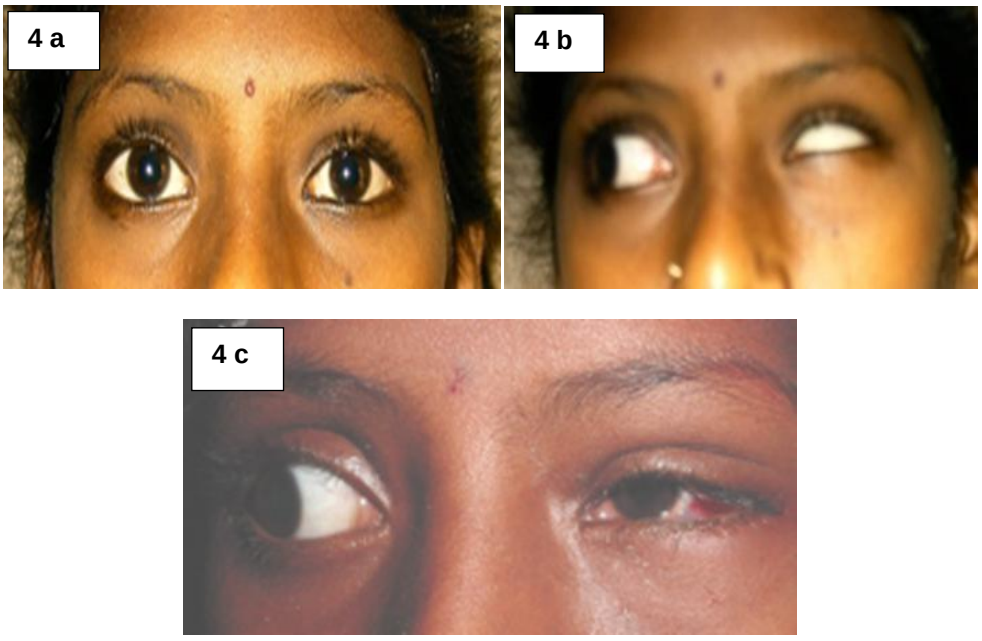

Fig. 4. Lateral rectus width 10mm: Recession only done. Pre op (a) primary gaze, (b) up shoot on adduction; Post op (c) no up/down shoot on adduction

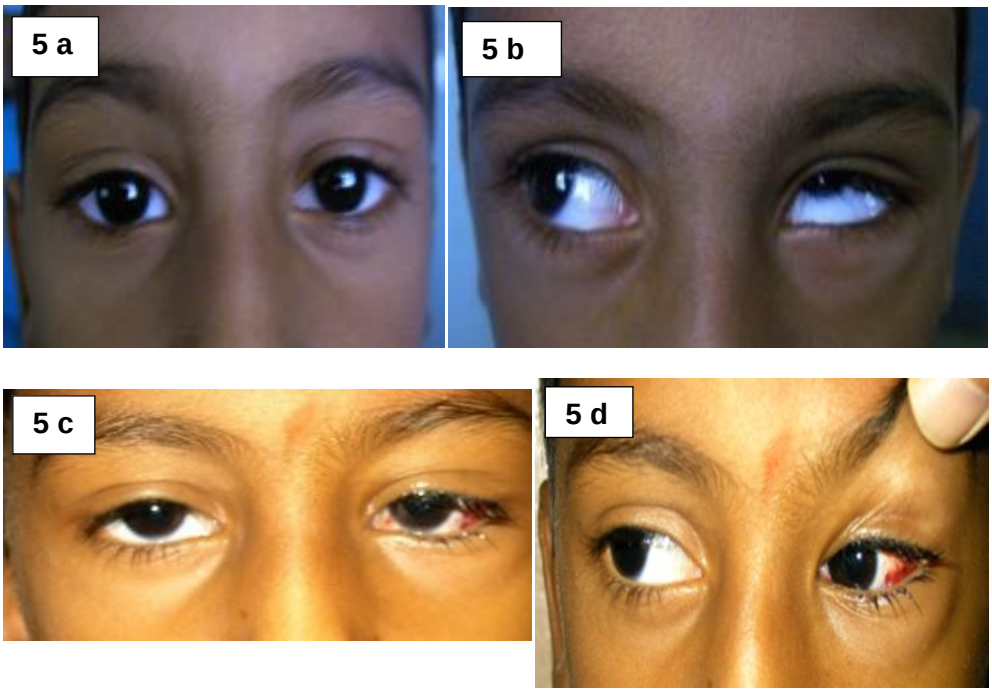

Fig. 5. Lateral rectus width 7mm: Lateral rectus recession + Y-splitting done. Pre op. (a) primary gaze, (b) upshoot on adduction; Post op. (c) primary position, (d) no upshoot on adduction

In these 5 patients, Y-splitting of LR was done in second sitting with good result. (Fig. 6). In all the 21 patients of group A, up/down shoot was almost nil after surgery. This group has 7 patients in whom width of lateral rectus was 7mm or less. Among 21 patients group B correction of up/down shoot was not satisfactory in 5 patients in whom width of lateral rectus at insertion was less than normal (7mm or less).

These results indicate that lateral rectus muscle Y splitting is essential apart from simple recession to reduce up/down shoot satisfactorily when lateral rectus insertion width was 7mm or less. Of the 12 patients with 7mm or less width, 7 underwent recession splitting with good result while 5 patients of group B in whom recession only was done had unsatisfactory result. The up/down shoots improved from 2-4 points to 0 points according to our scale of 0 to 4 (median 3.5).The correction of horizontal deviation was satisfactory. Mild hypertropia on down gaze was seen in 2 patients after recession splitting of lateral rectus, but both patients had orthotropia in primary position.

4. DISCUSSION

The goal of surgical correction of patients with Duane retraction syndrome is to relieve the abnormal head posture, to correct the deviation in primary position, to reduce the globe retraction, and to reduce the upshoot & down shoot.

Up-shoot and down-shoot in DRS patients are believed to be due to co contracture of the medial and lateral muscles and a taut LR muscle. Splitting of LR muscle lessens lateral slippage of the muscle and recession reduces of torque [19]. The effectiveness of Y-splitting depends on the restriction created by each of the two arms of the Y. As the eye looks above the midline in adducted position, the upper arm of Y rotates over the globe and the lower arm of Y, placed under further tension, contracts and prevents the eye from slipping upward and vice versa. Hence, the bifurcation of the muscle halves balances their position as the eye adducts. When globe retraction is present and the LR muscle is tight, it is essential to recess the LR muscle in addition to Y-splitting [20]. Rao et al. published a good result in 10 patients treated with Y-splitting and LR recession (with medial rectus recession in 6 patients) [16]. Lin reported equally favourable results for upshoot in 6 DRS patients, half with recession and other half with splitting of lateral rectus and with complication of hypertopia on down gaze with splitting [14].

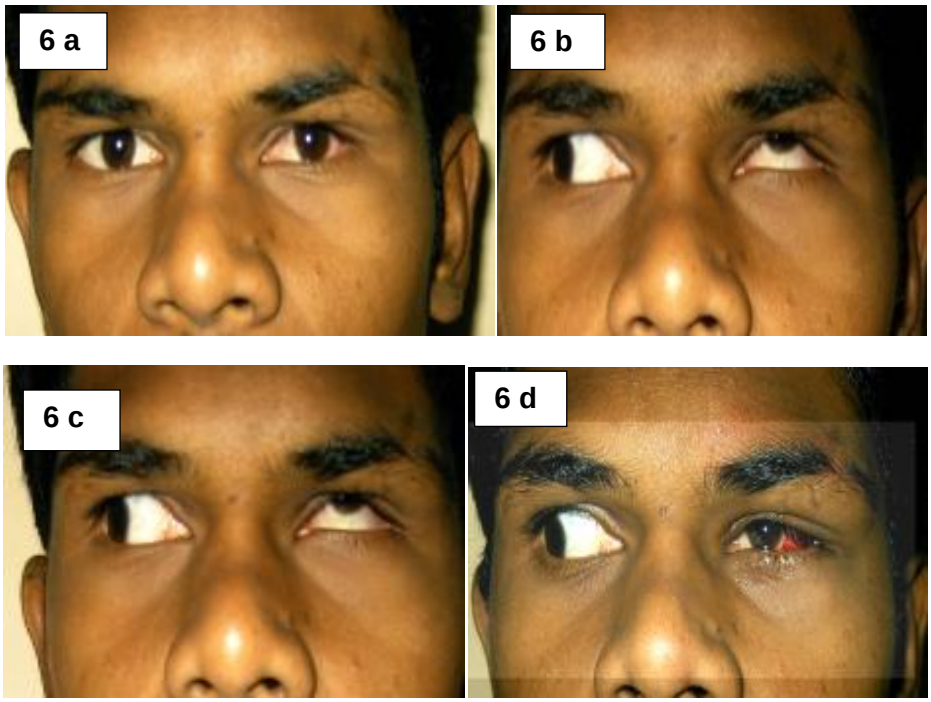

Fig. 6. Lateral rectus insertion width 6.5mm. Simple recession done. Pre op. (a) primary gaze, (b) upshot on adduction; Post op (c) upshoot not completely corrected, (d) lateral rectus Y-splitting additional surgery done -- upshoot corrected on adduction

Farid in his study concluded that combined Y-split recession procedure attained a more significant improvement of upshoot and down shoot but with higher incidence of postoperative complications [15]. Rogers and Bremer achieved marked decrease in up- and down-shoots in a series of 5 patients undergoing Y-splitting of LR muscle without recession [21]. Sukhija et al. reported 7 patients treated with Y-splitting and recession versus 8 patients treated with anchoring of the LR muscles to the lateral palpebral ligament and showed comparative results [22].

The success rate with recession splitting of lateral rectus was 100% as compared to 76.19% with simple recession (Fig. 7). In group A patients, preoperatively 5 cases had grade 4 upshoot, 12 cases grade 3 upshoot, and 4 cases grade 2 upshoot; while postoperatively all 21 cases had 0 grade upshoot. In group B patients, preoperatively 13 cases had grade 4 upshoot, 6 cases grade 3 upshoot, 1 case grade 2, and 1 case grade 1; while post operatively 1 case had grade 4 upshoot, 4 cases grade 3 upshoot, and 16 cases grade 0 upshoot. In our study all patients showed satisfactory improvement for up/down shoot except 5 of total 42 patients. After group wise analysis it was found that in all the 21 patients of group A, up/down shoot was nil after surgery. This group has 7 patients in whom the width of lateral rectus muscle was 7mm or less. Among the 21 patients of group B, the correction of up/down shoot was satisfactory in all except in 5 patients in whom the width of lateral rectus at insertion was 7mm or less. With all these observations it may presumed that narrow insertional width of lateral rectus may be responsible for up/down shoot in the process of leash effect. Simple recession will weaken the muscle, but the insertional width remains same which will not correct up/down shoot. If we increase the insertional width at the new insertion site, it will reduce the up/down shoot. Y splitting will increase the width at new insertion site and help in preventing slippage of the globe up or down. So splitting with recession of lateral rectus result in good correction of up/down shoot in situations where insertional width is narrow (7mm or less).

Another observation is that in 12 patients with 7mm or less width of lateral rectus, 8 patients underwent splitting with recession with good result while 5 patients of group B in whom recession alone was performed had unsatisfactory result.

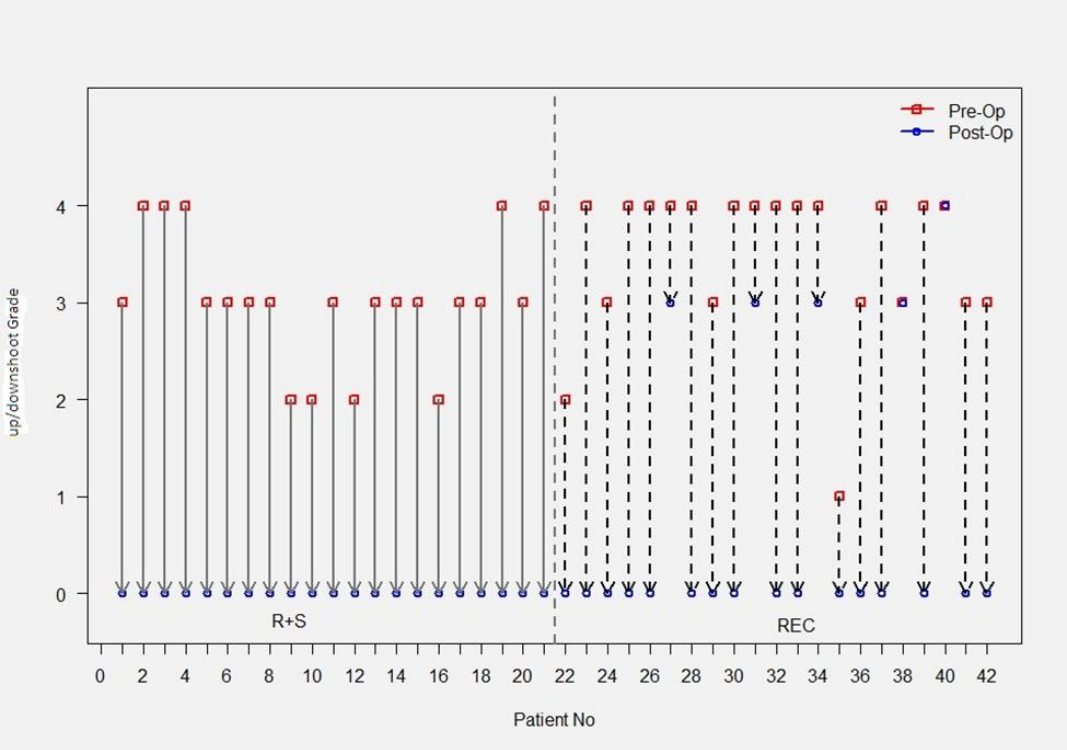

Fig. 7. Arrow plotting showing change in up/down shoot from preop (red) to postop (blue) in two groups. R+S=recession and splitting of lateral rectus, REC= recession of lateral rectus

These 5 patients had lateral rectus y- splitting in addition to already done recession again as second operation, which resulted in the improvement of upshoot. This shows that splitting of lateral rectus to increase insertional width is essential in addition to recession in patients with 7 mm or less lateral rectus insertion to correct the up/down shoot.

5. CONCLUSION

1) Recession of lateral rectus muscle with Y-splitting is a more effective (100%). procedure than simple recession (76.2%) in correcting significant upshoot and down shoot in Duane retraction syndrome patients.

2) Simple recession is not successful in significant proportion of cases (23.8%). In DRS with up/down shoot, lateral rectus insertion width is less than normal (7mm or less) in significant number of cases (28.5%). It is advised to measure lateral rectus width at insertion regularly intraoperatively to decide about splitting of lateral rectus.

3) Lateral rectus muscle Y- splitting is not necessary in DRS with upshoot / down shoot if the width of the muscle at insertion is within normal limits (8-12mm). In these cases simple recession of lateral rectus is enough.

4) The width of lateral rectus is one of the major factors in deciding the type of surgery (simple recession or with splitting of lateral. rectus) for correction of up/down shoot in DRS.

5) Since the number of patients is small (only 42) further study with large number of patients will be able to confirm our recommendations.

DISCLAIMER

The products used for this research are commonly and predominantly used products in our area of research and country. There is absolutely no conflict of interest between the authors and producers of the products because we do not intend to use these products as an avenue for any litigation but for the advancement of knowledge. Also, the research was not funded by the producing company; rather it was funded by personal efforts of the authors.

CONSENT AND ETHICAL APPROVAL

This study was approved by the Ethics committees of Osmania Medical College,

Hyderabad. Human patients sample collection strictly followed the ethical principles of the Declaration of Helsinki. Written informed consent was obtained from all patients for the surgery and for including in the study.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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