·Clinical Research·

Surgical outcomes for unilateral superior oblique palsy in Chinese population: a retrospective study

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Abstract

• AIM: To evaluate the outcome after surgery for unilateral superior oblique (SO) palsy in Chinese.

• METHODS: The medical records of 39 patients that underwent surgery for unilateral SO palsy between January 2003 and December 2012 at Caritas Medical Centre, Hong Kong, were retrospectively reviewed. All surgeries were performed by a single surgeon. Pre-operative assessments for vertical deviation, cyclo-deviation, and Knapp´s classification were obtained to determine the nature and degree of surgical correction. Vertical deviation was measured at 1wk; 1, 6mo and on last follow-up day post-operatively. Cyclo-deviation was measured on last follow-up day post-operatively.

• RESULTS: During the 10y period, 39 subjects were recruited. The most common etiology was congenital (94.9%). Knapp´s Type III (66.7%) and Type I (12.8%) classifications were the most common subtypes. To treat SO palsy, the most common surgical procedures were: isolated inferior oblique (IO) anteriorization (41.0%), isolated IO myectomy (10.3%), and isolated IO recession (10.3%). At 3.5 ±2.1y post-operatively, the vertical deviation was significantly reduced (15.1±6.2 PD versus 0.5±1.4 PD, *P*<0.0001) without significant improvement in cyclo-deviation (*P*=0.5). Initial vertical deviation was correlated with cyclo-torsion (*r*=0.4, *P*=0.007). Those with over-correction had greater initial vertical deviation (19.4±7.2 PD versus 13.2±4.3 PD, *P*=0.003). After a single operation, 84.6% of subjects achieved a vertical deviation within ±3 PD.

• CONCLUSION: The majority of subjects achieved corrected vertical deviation after a single surgery although there was no improvement in cyclo-deviation. Those with over-correction of primary position deviation had greater preoperative vertical deviation and it may be related to simultaneous multiple muscle surgery.

• KEYWORDS: superior oblique palsy; vertical deviation; surgery; diplopia
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INTRODUCTION

The trochlear nerve (4th cranial nerve) innervates the superior oblique (SO) muscle. SO palsy may cause not only vertical deviation, but also cyclo-deviation. Patients with unilateral SO palsy may present with hyper-deviation of the affected eye and abnormal head postures including head tilt and face turn to the contralateral side [1]. The diagnosis of SO palsy can be confirmed during clinical examination with a positive Park-Bielschowsky 3-step test (3-step test). The presence of a V pattern esotropia with chin elevation and reverse hypertropia during 3-step test should alert the clinician of the possibility of a bilateral SO palsy and confirmed with large excyclotorsion (>15 PD). Fundus examination for the foveal reflex can help to detect cyclo-deviation. The degree of cyclo-deviation can be measured by a double Maddox rod test while degree of vertical deviation can be measured by the alternate prism cover test in the 9 cardinal positions of gaze[2,3].

Patients with SO palsy presenting with intolerable vertical diplopia, torsional diplopia, or significant torticollis are indicated for treatment. The treatment of SO palsy is primarily surgical despite a few reports of success using prisms to alleviate the symptoms of vertical diplopia[4]. There are several surgical strategies for the correction of SO palsy with significant hypertropia. The surgical approaches for correction of vertical component depends on multiple considerations including: the Knapp's classifications, modified with deviation in primary gaze, the grading of inferior oblique (IO) overaction, the degree of SO underaction, presence of superior rectus (SR) contracture, pattern of commitance as well as the degree of the SO tendon laxity [5-7]. Most of the cases without SO tendon laxity can be successful treated by weakening of ipsilateral IO muscle [6]. For those presenting with large primary deviations >15-20 PD, it is recommended to perform a 2 or 3-muscle surgery [8,9]. Patients with torsional diplopia but without significant vertical deviation in the primary position can be treated with the Harada-Ito procedure with a success rate of 73% [10-12]. Appropriate and precise pre-operative and intra-operative assessment is the key to successful surgical correction.

The aim of this study was evaluate the surgical outcome for the treatment of unilateral SO palsy over a 10y period in a Chinese population.

SUBJECTS AND METHODS

The study was approved by Clinical Research Ethics Committee of the Hospital Authority of Hong Kong. The study was conducted in accordance with the Declaration of Helsinki. Informed consent was obtained from the patient's parents for the release and publication of clinical photos. The authors declare no financial or proprietary interests. This was a retrospective study conducted at Caritas Medical Centre, Hong Kong Special Administrative Region, China, between January 2003 and December 2012. The medical records of all patients that underwent surgical treatment for unilateral SO palsy during the study period were reviewed. All operations were performed by a single pediatric ophthalmologist (Tam VTY). All subject patients were diagnosed based on clinical examinations consisting of a positive 3-step test, evaluation of extraocular movements, as well as forced duction and force generation tests as further described below:

1) 3-step test combined with cover and uncover test was used to isolate the palsied muscle by observing the vertical deviation in primary gaze, left and right gaze, as well as right and left head tilt. For a subject with right SO palsy, a positive 3-step test would show a right hypertropia at primary position which is increased on left gaze and right head tilt.

2) Extraocular movement was performed with a fixation target to identify the underaction of the SO muscle and overaction of ipsilateral IO.

3) Forced duction test was performed under topical anaesthesia. The patient was instructed to move to a direction of limited movement, then the limbal conjunctiva was grasped firmly with a toothed forceps, the examiner would then further move the eyeball to see whether there was any mechanical restriction.

4) Forced generation test was performed under topical anaesthesia to assess the ability of a muscle to move the eye against a resisting force with examiner holding a toothed forceps placed on a limbal conjunctiva.

All patients underwent a pre-operative and post-operative orthoptic assessment by a single orthoptist. Assessments included: 3-steps-test, an alternate prism cover test measured at the 9 cardinal positions of gaze to identify the variation of deviations and to classify the palsy as per Knapp's classifications and a double Maddox rod test to detect and measure cyclo-deviation; presence of facial asymmetry and abnormal head posture [5]. Subjective improvements of abnormal head posture were assessed by parents and an orthoptist. Patients with less than 6mo post-operative follow-up were excluded.

Surgical Technique Surgical procedures were selected according to Knapp's subtypes, based on pre-operative measurement as well as intra-operative SO traction to detect for SO tendon laxity[7]. The selection of the operated muscles were based on previously reported recommendations [9,10] (Figure 1).

All surgeries were performed under general anesthesia. A forniceal conjunctival incision technique was used. For IO procedures, 8 mm post-limbal incision was made in the infero-temporal region. IO muscle was weakened with either a myectomy, recession, or anteriorization. IO myectomy was performed by removal of 6-8 mm segment of the intracapsular portion of the muscle. IO recession was performed in the manner described by Park's with anchoring the IO to Park's point (3 mm posterior and 2 mm lateral to temporal edge of inferior rectus (IR) insertion which equals to 10 mm of recession) [13]. For graded IO anteriorization along the border of IR, the procedure was performed with same surgical technique as described by Wright[14], where the amount of anteriorization depended on the pre-operative severity of IO overaction. For SO tucking, an intra-operative SO tendon traction test was performed as described by Plager [15]. For SO tendon laxity ≥ grade III, the SO tendon was tucked with the Bishop tendon tucker and secured with 5-O Ethibond non-absorbable sutures. The amount of tucking (6 to 8 mm) depended on the tendon laxity as well as the amount of vertical deviation in lateral gaze opposite to the paretic muscle. Conjunctival wound was closed with interrupted 8-O Vicryl absorbable sutures. Post-operatively, patients were given topical medications (0.1% dexamethasone and 0.5% Chloramphenicol) for 1mo. All patients were followed on 1d; 1, 4wk; 6mo post-operative, and then annually.

Outcome Measures The following data was collected: age, gender, laterality, etiology, signs and symptoms, surgical approaches, pre-operative and post-operative vertical and cyclo-deviations as well as Knapp's classification of SO palsy. The primary outcome measure was vertical deviation at 1wk; 1, 6mo and on the last follow-up day post- operatively. The secondary outcomes were post-operative cyclo-deviation and improvements in symptoms on last follow-up.

Statistics and Definitions The population was confirmed to be of Gausian distribution *via* the D'agostino&Pearson omnibus normality test. A Repeated Measures ANOVA with

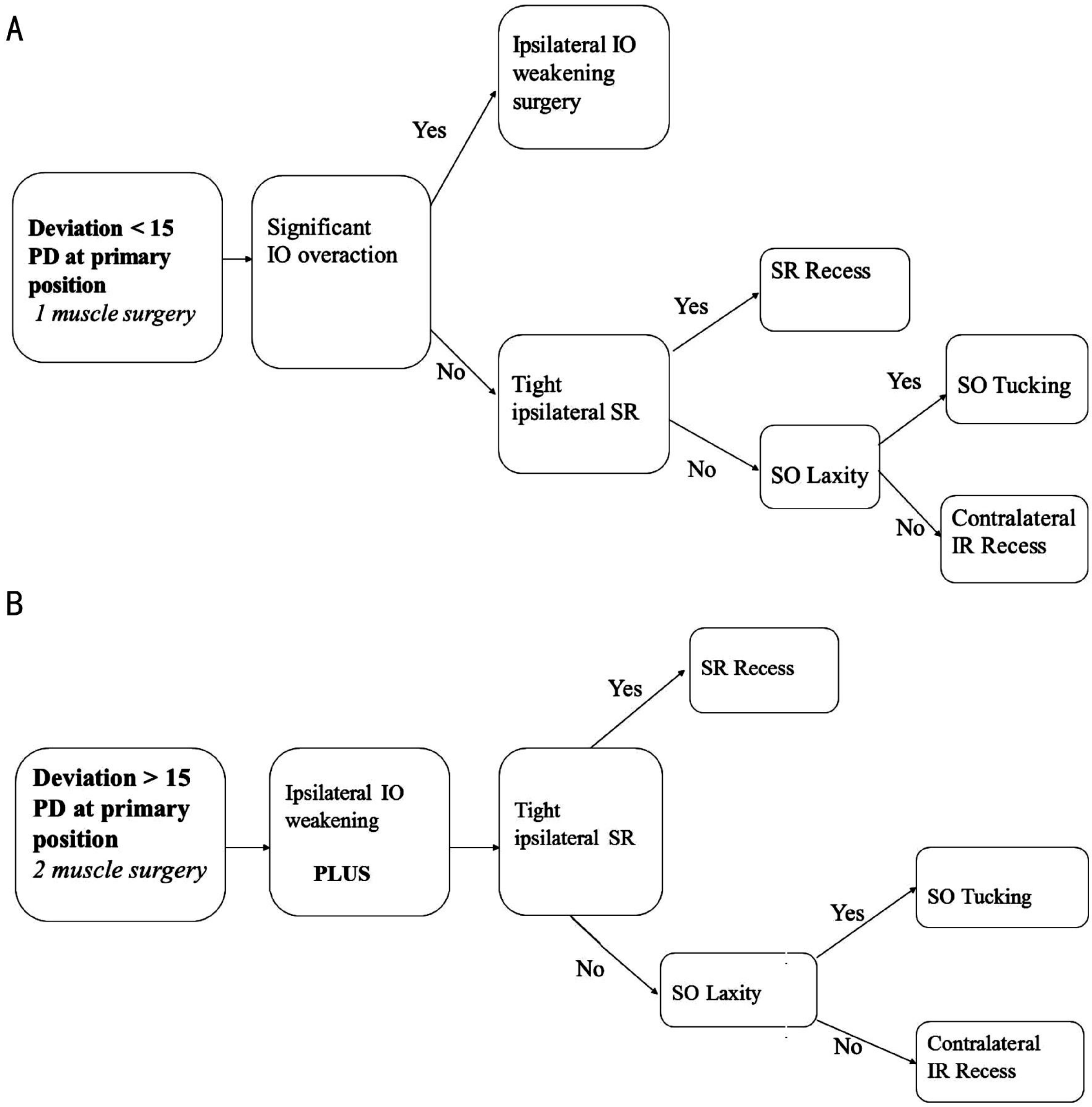

Figure 1 Flow chart for operational procedures based on vertical deviation on presentation A: Smaller deviation; B: Larger deviation.

Bonferroni's Multiple Comparison Test was used to analyze the changes in vertical deviation during the pre-operatively and post-operatively time intervals. Differences in pre-operative and post-operative vertical angle between male and female subjects were also compared using the Repeated Measures ANOVA with Bonferroni's Multiple Comparison Test where appropriate. Differences in pre-operative and post-operative cyclo-deviation were analyzed using a paired t-test. Correlation between pre-operative vertical deviation and cyclo-deviation was performed using the Pearson correlation coefficient. Correlation between the time to treatment versus the final vertical deviation was performed using the Pearson correlation coefficient. Absolute success was defined as vertical deviation within ±3 PD in primary gaze after a single operation. Qualified success was defined as correction of vertical deviation within ±3 PD in primary gaze after additional operations. For those with slight over-correction (vertical deviation between 0 to -3 PD on last follow-up) their vertical deviation on presentation was compared to those with vertical deviation between 0 to 3 PD on last follow-up using the t-test. All means were expressed as means±standard deviation and statistical significance was considered for $P<0.05$.

RESULTS

During the 10y period, 45 patients with unilateral SO palsy underwent surgical correction. Six patients with incomplete medical records were excluded. Thirty-nine cases were reviewed including 53.8% male and 46.2% female patients with a mean age of 15.8±18.1y. There were 56.4% right eyes and 43.6% left eyes. The common presenting signs and symptoms included abnormal head posture (head tilt and face turn), facial asymmetry, ocular misalignment, and diplopia. The most common etiology was congenital (94.9%) followed by trauma (2.6%), and stroke (2.6%). Knapp's type III disease was the most common (66.7%), following by type I (12.8%), type IV (10.3%), type II (7.7%), and type V (2.5%) (Table 1). The distribution of surgical procedures performed are summarized in Table 2.

Table 1 Demographic data *n* (%)

Demographic data	*n*=39
Age	
Mean (a)	15.8±18.1
Sex	
M	21 (53.8)
F	18 (46.2)
Laterality	
Right eye	22 (56.4)
Left eye	17 (43.6)
Signs and symptoms	
Abnormal head posture	32 (82.1)
Facial asymmetry	7 (51.3)
Ocular misalignment	19 (48.7)
Diplopia	5 (12.8)
Etiology	
Congenital	37 (94.9)
Trauma	1 (2.6)
Stroke	1 (2.6)
Knapp's classification	
Type I	5 (12.8)
Type II	3 (7.7)
Type III	26 (66.7)
Type IV	4 (10.3)
Type V	1 (2.5)

Table 2 Surgical procedures for the treatment of unilateral SO palsy *n* (%)

Surgical procedure	*n*=39
Isolated IO anteriorization	16 (41.0)
Isolated IO myectomy	4 (10.3)
Isolated IO recess	4 (10.3)
IO anteriorization+contralateral IR recess	5 (12.8)
IO anteriorization/myectomy+ ipsilateral SR recession	3 (7.7)
SO tucking+IO anteriorization	1 (2.6)
SO tuck+contralateral IR recession	1 (2.6)
SO tucking	3 (7.7)
IR recession	2 (5.1)
Harada-Ito	2 (5.1)

SO: Superior oblique; IO: Inferior oblique; IR: Inferior rectus; SR: Superior rectus.

The mean vertical deviation at presentation was 15.1±6.2 PD. The post-operative deviations were 0.6±1.2 PD at 1wk, 0.6±1.7 PD at 1mo, and 0.6±1.5 PD at 6mo, and 0.5±1.4 PD on last follow-up at 3.5±2.1y post-operatively. The vertical deviation was significantly reduced at all post-operative time points when compared to the pre-operative vertical deviation (all *P*<0.0001, Figure 2). There was no difference between the pre-operative and post-operative vertical deviation among male and female subjects (both *P*>0.05). There were significant reductions in the final post-operative vertical angle compared to pre-operatively in both male (14.8±5.2 *vs* 0.6±1.4 PD) and female (16.0±7.3 *vs* 0.5±1.4 PD) subjects (both *P*<0.0001).The mean pre-operative and post-operative cyclo-deviation was 1.3±0.8 PD and 1.2±0.8 PD at 3.5±2.1y post-operatively (*P*=0.5). There was a positive and significant

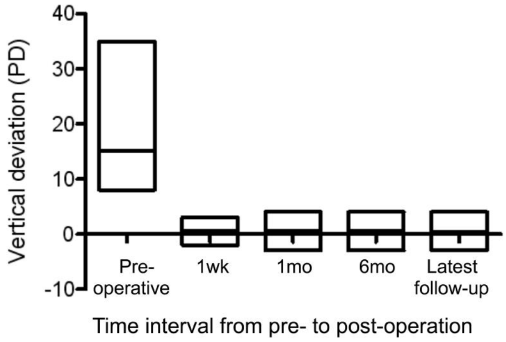

Figure 2 Mean±standard deviation of vertical deviations at various time intervals before and after operation.

correlation between the angle of vertical deviation and cyclo-torsion (*r*=0.4, *P*=0.007) on presentation. Those that had a slight over-correction (vertical deviation between 0 to -3 PD) on last follow-up had a greater vertical deviation on presentation compared to those that had a vertical deviation correction between 0 to 3 PD on last follow-up (19.4±7.2 PD *vs* 13.2±4.3 PD, *P*=0.003). There was no correlation between the time to treatment versus the final vertical deviation (*r*=0.05, *P*=0.7).

At 3.5±2.1y post-operatively, 78% of those with abnormal head posture had improvements. Most of patients were taken preoperative and postoperative photos to document the abnormal head postures and its improvement.

Ocular misalignment improved in 89.4% and 84.6% achieved absolute success with correction of vertical deviation after the first operation. Qualified success was improved to 94.8% after a second Harada-Ito procedure operation for 2 patients with persistent torsional diplopia.

Nine subjects (23.1%) in our series had pre-operative vertical deviation ≥20 PD. Among them, 7 had 2-muscle surgery and 2 had isolated SO tucking. For those with 2-muscle surgery, all achieved a final vertical deviation of ±3 PD. The other 2 patients who underwent isolated SO tucking, both of them had grade III SO laxity. The first patient had pre-operatively vertical deviation of +20 PD at primary position and excyclo-deviation of +2 PD. At 1wk post-operatively, there were improvements in both the vertical deviation (+4 PD) and excyclo-deviation (+1 PD); on last follow-up, the vertical deviation was +3 PD. For the second patient had pre-operatively vertical deviation of +25 PD at primary position and excyclotorsion of +3 PD. At 1wk post-operatively, the vertical deviation improved to +6 PD and the excyclotorsion improved to +2 PD; the vertical deviation was +5 PD on last follow up.

DISCUSSION

SO palsy can be congenital or caused by trauma, ischemic, demyelinating or inflammatory diseases. Even for congenital cases, patients may still present very late in life only when their fusional amplitude deteriorates [16]. Majority of strabismologists diagnosed congenital SO palsy based on a history of ocular torticollis since early childhood, increased vertical fusional amplitudes and absence of torsional diplopia.

In our series, 11 cases (29.7%) of congenital SO palsy presented to our centre after the age of 10.

Thirty-seven (94.8%) of our cases were congenital in nature. This proportion is much higher than the previously published data. Von Noorden and Campos [11] reviewed 270 cases with SO palsy and only 39.5% were congenital. Ellis *et al*[17] reported 76.9% of subjects belonging to congenital SO palsy. We postulate that the higher proportion of congenital cases in our series may be attributed to the fewer cases of trauma in our locality (sports or road traffic accident related).

In our study, the absolute surgical success was 84.6% (defined as vertical deviation within ±3 PD in primary gaze after a single operation) and the qualified surgical success was 94.8% (defined as correction of vertical deviation within ±3 PD in primary gaze after additional operations). In a small series of surgical management of 13 congenital SO palsy by Kekunnaya and Isenberg [18], their surgical success (defined as hypertropia <5 PD) was 69.0% (9/13, CI: 42.0% -88.0%). Similarly, in 39 Chinese patients with congenital SO palsy managed by Huang and Yu[19], 72.0% of cases were cured while 26% patients had improvements. In another Chinese study on the surgical management of 69 cases with unilateral SO palsy by Wang *et al* [20], 33.3% of patients were cured and 65.3% had improvements. Furthermore, in Peng *et al*'s[21] series of 112 Chinese patients with congenital SO palsy, the surgical success rate was ranged from 80.0% (anterior transposition of IO combined with contralateral IR recession) to 88.9% (anterior transposition of IO). The definitions for surgical success in managing SO (trochlear nerve) palsy varies among different studies with different cohort characteristics and pre-operative assessment methods, thus, it is often difficult to make direct comparisons [6,8,18-21]. For example, in Wang *et al*'s [20] study, pre-operative assessment was based on the degrees of the overaction of the IO muscle and the extent of vertical deviation in primary position where as in our case series, pre-operative assessment was based on Knapp's classifications.

Knapp's type III was the most frequently occurring subtype (66.7%) in our series, followed by type I (12.8%) and IV (10.3%), which is in line with the series reported by Bagheri *et al* [22] where type III (42.5%) was also the most common. However, Von Noorden and Campos[11] have reported type II (31%) to be the most common in their series.

Twenty-five subjects (64.1%) in our series had an isolated IO weakening procedure. All of whom belonged to Knapp's type I or III classification with vertical deviation <15 PD in primary position. The mean vertical deviation in our population at presentation was 15.1 ±6.2 PD. Eight-five percent of our population achieved a vertical deviation correction to within ±3 PD of primary gaze after a single surgery. Our surgical results were comparable with those of Hatz *et al* [23] that also recommended the use of isolated IO weakening procedure for the treatment of SO palsy up to 15 PD of vertical deviation in primary position.

From our experience, isolated SO tucking for large angle deviations (≥20 PD) could only achieve a success rate of 50% which was in agreement with Bhola *et al* [24] that concluded isolated SO tucking may not be sufficient to correct hypertropia greater than 15 PD.

Nejad *et al* [8] reported that only 14% of their patients achieved successful outcome after single muscle surgery for large vertical deviation whereas 58% had satisfactory result with multiple-muscle surgery. They recommended multiple-muscle surgery in SO palsy patients with large hypertropia in primary position. Combined 2-muscle surgery should be reserved for those with larger vertical deviations.

Five cases in our series (12.8%) underwent either isolated SO tucking or SO tucking combined to surgery on another muscle. Three patients (60%) achieved absolute success, 1 case developed an iatrogenic Brown's syndrome with limitation of elevation in adducted, although it was asymptomatic. The decision of SO tucking should be based on the intra-operatively SO tendon traction test to identify any significant SO laxity rather than solely on the amount of vertical deviation at primary position[24-26]. Iatrogenic Brown's syndrome is a well known post-operative complication of the SO tucking procedure. In a series by Durnian and Marsh[26], 10 out of 75 subjects developed iatrogenic Brown's syndrome but all were asymptomatic. Saunders *et al*[27] also commented that mild to moderate post-operative Brown syndrome is not undesired if it can be compensated for by long-term satisfactory outcomes.

Managing SO palsy is challenging for strabismus surgeons due to the wide range of pre-operative and intra-operative considerations. Early operation is recommended for paediatric patients with congenital etiology once the diagnosis is confirmed in order to achieve better cosmetic and functional outcome. Lau *et al* [28] showed that patients with congenital SO palsy operated at older ages are more likely to have residual torticollis, related to the underlying tightness of the sternocleidomastoid muscles. In our series consisting primarily of congenital SO palsy, for those that underwent operation under the age of 10, 80% (8/10) had their abnormal head posture corrected. For older patients, only 40% (10/25) resulted in resolution of abnormal head posture.

There was a significant reduction in the vertical deviation at all time points post-operatively, suggesting an immediate correction as early as 1wk post-operatively and consistently maintained for up to at 3.5±2.1y post-operatively. However, despite the correction in vertical deviation, there was no significant improvement in cyclo-deviation (P =0.5). Furthermore, a greater vertical deviation on presentation was significantly correlated with a greater cyclodviation (r=0.4, P=0.007). This is in contrast to the findings of Wei *et al*[29] and Gräf and Weihs[30] that reported a significant reduction in cyclovertical deviations after IO weakening procedures with or without SO tucking. This difference can be explained by the inconsistencies and variations of subjective assessment of cyclo-deviation as described by Mai *et al*[31]. Our study was limited by the absence of objective assessment of cyclo-devation such as with standard fundus photos as described by Wei *et al*[29].

Although absolute success was defined as a residual deviation within ±3 PD, 23.1% (9/39) of subjects had a post-operative deviation <0 PD suggesting a slight over-correction. When comparing their initial deviation to those that achieved a post-operative deviation of 0 to 3 PD, it was found that the over-correction group had a statistically greater vertical deviation on presentation (P=0.003). On the other hand, Nejad *et al*[8] reported that under-corrections were more frequent in those with an initial vertical deviation >20 PD at primary position. We postulate that these findings suggest that those with large angles on presentation have greater angle variability making pre-operative assessments more difficult, requiring multiple muscle operations, and hence greater post-operatively inconsistencies.

In summary, the majority of congenital SO palsies belonging to Knapp's type I or III achieved a post-operative vertical deviation within ±3 PD of primary gaze after a single 1-muslce surgery although there was no significant improvement in cyclotorsion. A larger initial vertical deviation was correlated to a larger initial cyclo-torsion, as well to over-correction after surgical treatment.

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