

## Surgical treatment of superior oblique palsy: Predictors of outcome

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**Purpose:** The purpose of this study was to evaluate the incidence and outcome of surgically treated superior oblique palsy (SOP) and the factors involved in its resolution. **Methods:** We performed a retrospective study of 76 patients who underwent surgery for SOP. We recorded data from the physical examination and the number and type of procedures performed. Favorable outcome was defined as resolution of or improvement in torticollis ( $\leq 5^\circ$ ) and diplopia in primary position (PP) and downgaze or as vertical deviation (VD)  $< 5$  prism diopters (pd) in PP and 10 pd in the oblique diagnostic position. **Results:** Mean age was 33.12 years. Congenital SOP was the most frequent type (65.8%). Mean preoperative VD was  $15.89 \pm 9.94$  pd, decreasing to  $3.07 \pm 4.36$  pd after surgery. Associated horizontal deviation was recorded in 51.32% of cases. The mean number of procedures was  $1.37 \pm 0.62$  (range 1–4), with 69.7% of patients requiring only one procedure. The mean number of muscles operated on was  $1.96 \pm 1.01$  (inferior oblique being the most frequent). A greater reduction in VD after surgery was observed in patients with congenital SOP ( $P = 0.04$ ). Although none of the factors evaluated influenced surgical outcome, amblyopic patients had a greater risk of reoperation ( $P = 0.04$ ). A favorable outcome was achieved in 75% of cases. Mean follow-up was 37.08 months. **Conclusion:** Congenital SOP was twice as frequent as acquired SOP and although surgery was successful in most cases, a greater reduction in VD was obtained in congenital cases. Amblyopia was identified as a risk factor for reoperation.

**Key words:** Acquired superior oblique palsy, amblyopia, congenital superior oblique palsy, multiple surgeries, surgical treatment

Superior oblique palsy (SOP) is the most frequent form of paralytic vertical strabismus and cranial nerve palsy.<sup>[1-3]</sup> It can be unilateral or bilateral and congenital or acquired.<sup>[4,5]</sup>

Treatment of SOP is complex and challenging, even for an ophthalmologist specializing in ocular motility. Treatment is mainly surgical and when indicated, aimed at resolving diplopia, torticollis, and vertical and torsional deviations.<sup>[6,7]</sup> Severe torticollis is the main indication for surgical treatment in children under the age of 5 years as the persistence of significant head tilt could lead to progressive facial asymmetry.<sup>[8]</sup> However, recent studies have associated the use of general anesthesia with brain injury, thus highlighting the importance of discussing the risks and benefits of early surgical treatment in SOP.<sup>[8]</sup>

Most published studies report favorable outcomes with inferior oblique muscle surgery; consequently, this muscle is the most frequently operated on.<sup>[6,7,9-12]</sup> However, some studies suggest that in significant ( $> 20$  prism diopters [pd]) vertical deviation (VD), two or more muscles would need to be operated on to achieve a favorable outcome with a low risk of postsurgical overcorrection. Furthermore, in such cases, the results improve after a second intervention.<sup>[13,14]</sup>

No consensus has been reached on the best combination of muscles to be operated on to yield optimal results and thus avoid reoperations in the treatment of SOP. A recommendation based on the current evidence is to individualize surgery

according to the deviation present in each of the 11 gaze positions (including the Bielschowsky maneuver) and to whether SOP is congenital or acquired, unilateral or bilateral.<sup>[2]</sup>

Limited evidence is available regarding the factors associated with successful surgery. For some authors, the presence of severe VD in the initial examination and the involvement of several muscles are associated with a higher rate of surgical overcorrection.<sup>[15]</sup> Other factors, such as age at the time of surgery, have been shown to have an impact both on the outcome of surgery and on the need for reoperation.<sup>[16,17]</sup>

The purpose of the present study was to evaluate the incidence and outcome of surgery to treat SOP and the factors involved in its resolution in the ocular motility section of a tertiary center.

## Methods

We performed a retrospective study of 76 patients with unilateral or bilateral SOP who underwent surgery in our institution between 2001 and 2015. All operations were performed by two surgeons (PM and PGL), each of whom had received 25 years' training in strabismus surgery. The study was performed in accordance with the Declaration of Helsinki and approved by the local Ethics Committee.

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Patients with a diagnosis of masked bilateral SOP were also included in the study. Patients who had been followed for <6 months and those who had been treated with botulinum toxin only were excluded from the study.

The diagnosis of congenital or idiopathic SOP was made based on available photographic evidence of torticollis since childhood and in the absence of other causes, such as history of trauma and associated diseases. Patients were diagnosed with acquired SOP when associated disorders were present and onset of symptoms was acute.

Each patient's medical history was reviewed to collect demographic data (e.g., age, gender, and etiology) and motor and sensory examination data (e.g., vertical and horizontal deviations in primary position [PP], fibrosis of the superior rectus [SR], VD with the Bielschowsky maneuver, and presence of diplopia) [Table 1].

The presence of torticollis was demonstrated by making the patients fix their gaze on the smallest optotype line that they could visualize with correction. Deviation in PP was assessed

**Table 1: Motor and sensory examination**

	Initial	Final
Vertical deviation (mean±SD)	15.89±9.94 pd	3.08±4.36 pd
Initial horizontal deviation (mean±SD)		
Esotropia	9.11±6.58 pd	
Exotropia	-5.4±3.24 pd	
Initial torticollis (mean±SD)	13.84°±7.15°	4.28°±4.87°
Initial Bielschowsky (mean±SD)	18.60±11.07 pd	3.60±4.80 pd
Inferior oblique overaction, <i>n</i> (%)		
0	1 (1.3)	30 (39.5)
+1	23 (30.7)	37 (48.7)
+2	28 (37.3)	9 (11.8)
+3	23 (30.7)	
Superior oblique underaction, <i>n</i> (%)		
0	3 (4.1)	47 (61.8)
-1	38 (51.4)	22 (28.9)
-2	29 (39.2)	6 (7.9)
-3	4 (5.4)	1 (1.3)
Initial diplopia, <i>n</i> (%)	47 (61.8)	20 (26.3)
Position of maximum diplopia at presentation, <i>n</i> (%)		
Supradextroversion	14 (31.8)	
Dextroversion	3 (6.8)	
Infradextroversion	6 (13.6)	
Supraversion	2 (4.5)	
Primary position	1 (2.3)	
Infraversion	3 (6.8)	
Supraleversion	4 (9.1)	
Levoversion	3 (6.8)	
Infraleversion	8 (18.2)	
Initial fusion (worth test), <i>n</i> (%)	47 (67.1)	
Initial stereopsis (TNO), <i>n</i> (%)	22 (31)	
Initial superior rectus fibrosis, <i>n</i> (%)	27 (35.5)	

SD: Standard deviation

using the alternate prism cover test. Overaction of the inferior oblique muscle was graded from +1 to +3, while underaction of the superior oblique muscle was graded from -1 to -3. Subjective excyclotorsion was evaluated using double Maddox rod testing and objective excyclotorsion by retinography after mydriasis with cyclopentolate 1% or tropicamide.

Amblyopia was defined as visual acuity <1/2 in one eye or a difference of two or more lines of the Snellen optotype between both eyes. Binocular vision and stereopsis were assessed using the Worth and TNO tests, respectively. A Lancaster screen test was performed before and after surgery in all patients who were able to cooperate.

Fibrosis of the SR secondary to SOP was defined as a limitation of the depression in abduction of the affected eye with a greater VD in that position and a difference of at least 15 pd in VD between both eyes in the Bielschowsky maneuver.

Our criteria for surgical treatment of intravenous (IV) nerve palsy included individualization of each case according to the cover test in the 11 positions of gaze, the position of maximum diplopia, and the results of the Lancaster test. Surgery was performed on one cyclovertical muscle for deviations ≤15 pd in PP, whereas two muscles were operated on simultaneously for deviations >15 pd.

Botulinum toxin (Botox®, Allergan, Inc., Irvine, California, USA) was used as a complementary treatment for symptomatic surgical under- and over-correction and as a first-line therapy for acquired SOP diagnosed <4 months previously or for acquired SOP diagnosed >4 months previously with VDs in PP <6 pd. Botulinum toxin was not administered to patients aged <18 years, owing to the inability to perform the technique under topical anesthesia with electromyographic identification of the inferior oblique and rectus muscles during elevation and depression of the eye. An injection of 2.5–5 IU was administered in the inferior rectus and/or inferior oblique muscles depending on the cover test result (11 positions) and Lancaster test findings.

Surgery was performed in acquired SOP of at least 6 months' duration and at the time of diagnosis in cases of decompensated congenital SOP.

Favorable outcome was defined as resolution or improvement of torticollis (≤5°) and diplopia in PP and downgaze or as a residual VD ≤5 pd in PP and 10 pd in the oblique diagnostic positions.

The statistical analysis was performed using IBM SPSS Statistics for Windows, Version 22.0 (IBM Corp., Armonk, New York, USA). The Kolmogorov–Smirnov test was applied to determine whether the variables in our sample followed a normal distribution. The *t*-test was used to compare normally distributed quantitative variables, and the Mann-Whitney test was performed to compare nonnormally distributed quantitative variables. Pearson's Chi-square test was used to compare qualitative variables. A logistic regression analysis of the surgical outcome-dependent variables and the number of procedures was carried out. A *P* < 0.05 was considered statistically significant.

## Results

The study population comprised 76 cases (60.5% males [46/72]), with a mean age of 33.12 ± 2.64 years (range 2–80). The left

eye was affected in 52.6% of cases, and only 5.3% presented bilateral SOP. The right eye was dominant in 51.5% of patients. The most frequent etiology was decompensated congenital or idiopathic SOP, which was present in 50 cases (65.8%). Acquired SOP (34.2%,  $n = 26$ ) was classified as traumatic (14.5%,  $n = 11$ ), iatrogenic (11.8%,  $n = 9$ ), neoplastic (3.9%,  $n = 3$ ), and vascular (3.9%,  $n = 3$ ).

Clinical data from the initial and final examinations are recorded in Table 1. The mean initial VD was  $15.89 \pm 9.94$  pd (range 0–45 pd). Associated horizontal deviation was present in 51.32% of patients, exotropia in 39.97% (mean  $\pm$  standard deviation [SD]:  $-5.4 \pm 3.24$  pd), and esotropia in 11.85% (mean  $\pm$  SD:  $9.11 \pm 6.58$  pd). Presurgical VD according to the Bielschowsky maneuver was  $18.60 \pm 11.07$  pd (range 0–50 pd). Postsurgical mean VD was  $3.07 \pm 4.36$  pd in PP and  $3.6 \pm 4.8$  pd with the Bielschowsky maneuver. SR fibrosis was present in 35.5% of patients. Although 94.73% presented with torticollis initially (mean  $\pm$  SD:  $13.84 \pm 7.15^\circ$ ), torticollis (mean  $\pm$  SD:  $4.27 \pm 4.87^\circ$ ) persisted at the end of the follow-up period in 55.26% of patients. Diplopia was recorded before surgery in 61.8% of cases. After treatment, diplopia was not observed in any of the gaze positions in 73.7% of patients. Subjective and objective excyclotorsion were present in 67.4% of cases (31/46) and 64% of cases (14/25) in which they were evaluated, respectively. Amblyopia was present in 14.5% of patients.

Information regarding the surgical procedures performed is available in Table 2. The mean number of procedures was  $1.37 \pm 0.62$  (range 1–4), with 69.7% of patients requiring only one procedure. Two interventions were needed in 25% of cases, and three and four procedures were performed in 3.9% and 1.3% of patients, respectively. The mean total number of muscles operated on was  $1.96 \pm 1.01$  (range 1–4). The Apt inferior oblique procedure was the most frequent approach in first interventions (30.3%), followed by combined SR recession and superior oblique resection (11.8%), Apt with inferior rectus recession (9.2%), and isolated inferior rectus recession (9.2%). The most frequent procedure was the Apt technique, which was performed in 53.9% of cases either alone or combined with another technique. In the cases that required a second intervention, inferior rectus recession was the most frequent (9.2%) [Fig. 1]. Topical anesthesia was used in 56.6% of procedures. Horizontal rectus surgery was also performed in only 5 cases. Botulinum toxin was administered in 22.4% of patients before surgery and in 17.1% after surgery.

The differences between congenital and acquired SOP are shown in Table 3. A greater reduction in VD was observed in congenital SOP ( $P = 0.04$ ) after surgery. Patients with acquired SOP received more injections of botulinum toxin and higher doses than patients with congenital SOP ( $P = 0.001$ ). Although none of the variables evaluated (age, torticollis, VD, etiology of palsy, presence of amblyopia, number of muscles operated on, type of anesthesia, and SR fibrosis) could be identified as a factor predicting good surgical outcomes in SOP [Table 4], amblyopic patients had a greater risk of reoperation, with 54.54% requiring more than 1 surgical procedure compared with 26.15% of nonamblyopic patients ( $P = 0.04$ ) [Table 5]. A favorable surgical outcome was achieved at the end of the study in 75% of cases, with a mean follow-up period from the last intervention of  $37.08 \pm 34.58$  months (range 6.02–132.83).

**Table 2: Surgical treatment and botulinum toxin (Botox) injection data**

Total number of Botox injections (mean $\pm$ SD)	1.5 $\pm$ 2.73
Total Botox dose (mean $\pm$ SD)	6.81 $\pm$ 11.91 IU
Presurgical Botox, $n$ (%)	17 (22.4)
Postsurgical Botox, $n$ (%)	13 (17.1)
Total number of procedures (mean $\pm$ SD)	1.37 $\pm$ 0.62
Total number of muscles operated on (mean $\pm$ SD)	1.96 $\pm$ 1.01
Type of anesthesia, $n$ (%)	
General	33 (43.4)
Topical	43 (56.6)
Number of muscles at first surgery (mean $\pm$ SD)	1.50 $\pm$ 0.70
Number of muscles at first surgery, $n$ (%)	76 (100)
1 muscle	45 (59.2)
2 muscles	26 (34.2)
3 muscles	3 (3.9)
4 muscles	2 (2.6)
Number of muscles during second procedure (mean $\pm$ SD)	1.36 $\pm$ 0.49
Number of muscles during second procedure, $n$ (%)	22 (28.9)
1 muscle	14 (63.6)
2 muscles	8 (36.4)
Number of muscles during third procedure, $n$ (%)	3 (3.94)
1 muscle	3 (100)
Number of muscles during fourth procedure, $n$ (%)	1 (1.31)
1 muscle	1 (100)

SD: Standard deviation

## Discussion

In the present study, we found that congenital SOP was twice as frequent as acquired SOP, thus raising the possibility that acquired SOP resolves more frequently after the administration of botulinum toxin or even spontaneously. In our sample, 66% of cases were congenital as reported elsewhere.<sup>[2]</sup> Traumatic cases were more common than other types of acquired cases as previously described.<sup>[2-4,18]</sup> Bilateral SOP was present in only 5.3% of our cohort, which is lower than percentages reported in other series.<sup>[19]</sup> This difference could be explained by the fact that bilateral cases are most frequently acquired and respond well to botulinum toxin alone;<sup>[4,5]</sup> therefore, they were excluded from our study.

Although VD, torticollis, and excyclotorsion are classic features of IV nerve palsy, some patients also present associated horizontal deviation (exotropia or esotropia). In our cohort, 51.32% of patients had horizontal strabismus, mainly exotropia, which did not require surgical treatment in most cases (only 6.57% [5/76] required surgery). Previous studies reported that associated horizontal strabismus was frequent (50%–60% of patients), both in congenital SOP and in acquired SOP,<sup>[2,13,20]</sup> and that surgery on the horizontal rectus muscles is only needed in deviations  $>8$  pd<sup>[20]</sup> or 15 pd<sup>[2,13]</sup> since most of the less severe cases could be resolved with surgery involving the cyclovertical muscles.

Outcome was favorable in 69.7% of cases with only one procedure, and in 59.2% of patients, only one muscle was operated on; in 34.2% of cases, it was necessary to operate on two muscles. At the end of the follow-up period, outcome was favorable in 75% of patients after one or more surgical interventions. The muscle most frequently operated on (54%) was the inferior oblique (either alone or in association with the contralateral inferior rectus or ipsilateral superior oblique).

Treatment of SOP is mainly surgical. However, the type and number of muscles to be operated on during the first procedure remain unclear. Hence, some authors recommend operating routinely on one muscle only for first procedures to avoid overcorrections and waiting before planning a second intervention.<sup>[17,21,22]</sup> Most published studies report techniques that involve more than one muscle, implicating the inferior oblique and/or superior oblique with the contralateral inferior

rectus or ipsilateral SR muscle, especially when SR fibrosis or contracture leads to greater VD.<sup>[7,23,24]</sup>

Reports from various series show that surgery is successful in 58%–95% of cases.<sup>[2,13,23–25]</sup> This difference in frequency could be explained by the different criteria for good surgical results used by each author as well as the clinical heterogeneity of samples. Inferior oblique recession yields worse results than myectomy in VD >15 pd.<sup>[6]</sup> Inferior oblique resection and anteropositioning yield good results in VD of 20–25 pd<sup>[11]</sup> as reported elsewhere.<sup>[13]</sup> However, isolated inferior oblique recession should only be used for VD <15 pd.<sup>[9]</sup>

Only a small percentage of patients received complementary treatment with botulinum toxin before and after surgery. Patients with acquired SOP required more injections and higher doses ( $P = 0.001$ ) than patients with congenital SOP. This observation could be explained by the fact that botulinum toxin was not indicated in patients younger than 18 although no differences were found between congenital and acquired cases with regard to age.

Reduction of VD with treatment was greater in congenital cases ( $P = 0.04$ ); however, we were unable to compare our observations owing to lack of data in the literature. SR fibrosis contracture, which was found to be more frequent and long-standing in congenital cases (40%) than acquired cases (26.9%), could be associated with a better response to surgery.<sup>[24]</sup> Amblyopia was found to be a risk factor for reoperation in SOP ( $P = 0.04$ ), thus highlighting the importance of treating it in pediatric patients before indicating early surgical interventions in congenital SOP. Other authors found that older age and greater vertical and torsional deviations were involved in the need for reoperation.<sup>[17,26,27]</sup> None of the other factors evaluated, such as age, torticollis, deviation, etiology of palsy, presence of amblyopia, number of muscles, type of anesthesia, and SR fibrosis, were found to affect the final outcome. According to our results, we recommend performing surgery on the inferior oblique for deviations  $\leq 15$  pd in PP, except in the fibrosis of SR, whereas the SR recession will be the procedure of choice. Two muscles must be operated on simultaneously for deviations >15 pd. The muscle combination

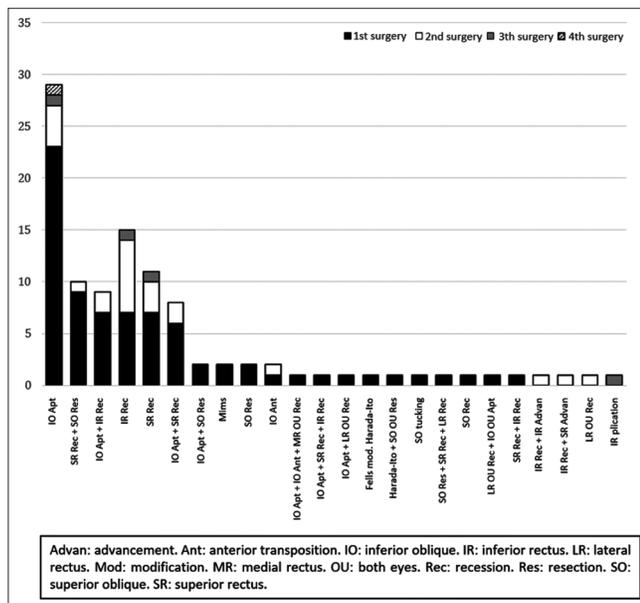


Figure 1: Types of procedures

Table 3: Differences between congenital and acquired superior oblique palsy

	Congenital (n=50), n (%)	Acquired (n=26), n (%)	P
Age	30.56±21.46	38.04±21.54	0.154
Sex (male/female)	29 (58)/21 (42)	17 (65.38)/9 (34.62)	0.532
Superior rectus fibrosis	20 (40)	7 (26.9)	0.258
Vertical deviation reduction	-14.38±9.75	-9.81±8.71	<b>0.048</b>
Bielschowsky reduction	-15.30±11.10	-14.04±12.30	0.666
Amblyopia	5 (10)	6 (23)	0.124
Torticollis	-10.14±6.833	-8.46±7.845	0.549
Number of procedures	1.34±0.55	1.42±0.75	0.859
Number of muscles	1.90±0.93	2.08±1.16	0.668
Botox before surgery	4 (8)	13 (50)	<b>0.001</b>
Botox after surgery	7 (14)	6 (23.07)	0.319
Number of Botox injections	0.88	2.69	<b>0.001</b>
Total dose of Botox	4.01±10.25	12.21±13.16	<b>0.001</b>
Favorable outcome	36 (72)	21 (80.76)	0.402

Statistical significance was set at <0.05. Statistically significant values are highlighted in bold

**Table 4: Predictive factors of surgical outcome**

	Favorable outcome, n (%)	Poor outcome, n (%)	P
Age	34.63±22.60	28.58±18.24	0.294
Initial vertical deviation	15.44±9.02	17.26±12.51	0.492
Amblyopia	7 (12.3)	4 (21.1)	0.347
Initial torticollis			
Range	36.15	45.55	0.099
Etiology			
Congenital	36 (63.2)	14 (73.3)	0.402
Acquired	21 (36.8)	5 (26.3)	
Number of muscles operated on			
Range	37.61	41.16	0.521
Superior rectus fibrosis	20 (35.1)	7 (36.8)	0.890
Type of anesthesia			
General	25 (43.9)	8 (42.1)	0.894
Topical	32 (56.1)	11 (57.9)	

Statistical significance was set at <0.05

**Table 5: Predictors of multiple procedures**

	One procedure	Two or more procedures	P
Age	34.70±21.12	29.48±22.86	0.092
Vertical deviation	15.66±11.09	16.43±6.76	0.644
Amblyopia	5 (9.43)	6 (26.08)	<b>0.040</b>
Torticollis	13.81±7.65	13.91±6.02	0.836
Etiology			
Congenital	35 (66.03)	15 (65.22)	0.697
Acquired	18 (33.97)	8 (34.78)	
Superiorrectus fibrosis	18 (33.96)	9 (39.13)	0.846
Type of anesthesia			
General	23 (43.40)	10 (43.48)	0.190
Topical	30 (56.60)	13 (56.52)	

Statistical significance was set at <0.05. Statistically significant values are highlighted in bold

will be the inferior oblique with the contralateral inferior rectus or ipsilateral SR muscle, depending on whether the deviation is greater in infraversion or supraversion, respectively.

The main limitation of our study is its retrospective design, which prevented us from recording specific data; for example, subjective torsion and objective torsion were not documented in all cases, thus making it difficult to evaluate the influence of these factors on the final outcome and the need for reoperation. However, our sample, which is of an appropriate size, included patients evaluated over a long period (14 years), with long follow-up times and standard surgical criteria for SOP even though the surgical approach had to be tailored to the patient.

## Conclusion

In a cohort of patients with SOP undergoing surgery, we found that idiopathic and congenital SOP were twice as frequent as acquired SOP, of which trauma was the most frequent type. Although favorable surgical results were obtained in most cases with only one procedure, approximately half the patients required surgery on more than one muscle, with the inferior

oblique muscle the most frequently operated on. We found that the change in VD after surgery was greater in congenital SOP and that acquired cases received higher doses and a greater number of botulinum toxin injections. Amblyopia was identified as a risk factor for reoperation, but none of the other variables evaluated had an impact on the outcome of surgery at the end of follow-up.

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## Conflicts of interest

There are no conflicts of interest.

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