Diplopia after strabismus surgery for adults with nondiplopic childhood-onset strabismus

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PURPOSE To describe frequency of postoperative diplopia after strabismus surgery in nondiplopic adults with childhood-onset strabismus and to report health-related quality-of-life (HRQOL) outcomes.

METHODS We prospectively enrolled 79 adults with no diplopia in any gaze who had childhood-onset strabismus and were scheduled for strabismus surgery. Diplopia was assessed preoperatively and at 6 weeks and 1 year postoperatively using a standardized diplopia questionnaire with 5 response options in 7 gaze positions. HRQOL was assessed using the Adult Strabismus-20 (AS-20) questionnaire, with self-perception, interactions, reading function, and general function domains.

RESULTS Constant diplopia in straight-ahead distance and reading gaze occurred in 1 patient (1% [95% CI, 0%-7%] at 6 weeks and 2% [95% CI, 0%-10%] at 1 year). Regarding the rate of any diplopia (including rarely) in any gaze, 15 of 78 patients (19%) reported diplopia at 6 weeks, of whom 13 had diplopia in straight-ahead distance gaze; 8 (10%), in reading gaze. At 1 year, 8 of 51 patients (16%) reported any diplopia (including rarely) in any gaze, of whom 7 had diplopia in straight-ahead distance gaze and 4 (8%) in reading gaze. Mean AS-20 scores improved at 1 year overall (by 32, 19, 14, and 15 points, resp., per domain) and for the 8 diplopic patients (by 21, 13, 16, and 11 points).

CONCLUSIONS In adults with nondiplopic strabismus, constant postoperative diplopia is rare, although the rate of intermittent diplopia is higher. Even when postoperative diplopia occurs, HRQOL often improves. (J AAPOS 2019;23:313.e1-5)

The benefits of strabismus surgery in adults are well established and include restoration of alignment, better visual function, improved self-perception, and improved interpersonal interactions.[1-3] In light of these potential benefits, many adults with childhood-onset strabismus are offered strabismus surgery. Nevertheless, some patients and providers have fears of new-onset postoperative diplopia and for that reason avoid surgery. The occurrence of new-onset postoperative diplopia in adults with nondiplopic childhood-onset strabismus is reportedly rare (0.8%-4%),[4-6] but its occurrence has not been systematically studied using a standardized patient-reported outcome measure.

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The Diplopia Questionnaire[7] was designed as a standardized patient-reported outcome measure for symptoms of diplopia to allow reporting of diplopia severity in 7 gaze positions. It allows for quantification of diplopia on a 0-100 scale. A standardized patient-reported measure has also been found to be more comprehensive than the patient history recorded in the medical record.[8] The purpose of this study was to describe the frequency of postoperative diplopia after strabismus surgery in adults with childhood-onset strabismus without preoperative diplopia and report health-related quality of life (HRQOL) outcomes of these patients.

Subjects and Methods

The procedures used in this study conformed to the Declaration of Helsinki and were approved by the Mayo Clinic Institutional Review Board.

We prospectively enrolled 79 nondiplopic adult patients undergoing strabismus surgery from March 15, 2010, to June 30, 2016, in the strabismus practice of one author (JMH). To be included, patients had to be at least 16 years of age at time of surgery, with strabismus onset before age 9. Patients also had to have completed a standardized diplopia questionnaire[7] and the Adult Strabismus-20 (AS-20)[9,10] questionnaire preoperatively and 6 weeks (window, 3 weeks to 5 months) and 1 year (window,

5 months to 2 years) postoperatively. To ensure that patients did not have diplopia before strabismus surgery, only those who answered "never" in all positions of gaze on the diplopia questionnaire preoperatively and not wearing prism were included. Patients with a history of intermittent exotropia, convergence insufficiency, and trochlear nerve palsy were excluded because of the difficulty defining age of onset for these conditions. Patients with primarily restrictive strabismus were also excluded, because these conditions are typically associated with diplopia and do not represent a common type of childhood-onset strabismus; however, patients with, for example, consecutive exotropia with a tight lateral rectus muscle causing mild limitation of adduction were included.

Data collected included demographics (age, sex, race), original (ie, childhood) diagnosis, presenting diagnosis as an adult, preoperative alignment, and postoperative alignment at 6 weeks and 1 year. All motor alignment measurements were made using the prism and alternate cover test.

Questionnaires

All patients completed a standardized diplopia questionnaire that allowed patient reporting of diplopia severity in 7 gaze positions.[7] Patients were asked whether over the course of the week they experienced diplopia for reading, straight-ahead distance gaze, down, right, left, up, and any other gaze position. For each gaze position, the patient could select 1 of 5 severities: never, rarely, sometimes, often, or always.

The AS-20 assesses HRQOL and functional vision in adults with strabismus[9] in four distinct domains: self-perception, interactions, reading function, and general function. Each domain is scored independently using Rasch scoring methods and converted to a score of 0-100 (worst to best HRQOL).[10]

Data Analysis

Postoperative diplopia was evaluated at 6 weeks and 1 year after surgery. We calculated the proportions of patients, with 95% confidence intervals, with (1) any diplopia in any gaze position, (2) any diplopia in straight-ahead distance gaze and/or reading gaze, and (3) constant diplopia in straight-ahead distance gaze and/or for reading.

To assess HRQOL, we calculated mean AS-20 scores for each patient for each domain at pre- and postoperative time points. Mean change in AS-20 scores and 95% confidence intervals were calculated at 6 weeks and 1 year for each domain for both the overall cohort and for the patients with postoperative diplopia. *P* values were calculated using signed-rank tests.

Results

Of the 79 patients enrolled, mean age at time of surgery was 40 ± 18 years (range, 16-82 years); 47 (59%) were female, and 76 (96%) were white. The original childhood diagnosis was esotropia in 65 (82%), exotropia in 12 (15%), hypertropia in 1 (1%), and original diagnosis unclear for 1 (1%). The presenting preoperative diagnosis was exotropia in 47 (59%), esotropia in 27 (34%), and hypertropia in 5 (6%). Details of childhood and presenting strabismus types are presented in Table 1. Preoperative data were obtained a median of 1 day before surgery (range, 1-13 days). Postoperative 6-week data were obtained at a median of 7 weeks after surgery (range, 4-18 weeks), and postoperative 1-year data were obtained at a median of 53 weeks after surgery (range, 23-93 weeks).

Table 1. Childhood and presenting strabismus diagnoses in 79 nondiplopic adults undergoing surgery for childhood-onset strabismus

Diagnosis	No. (%)
Childhood diagnosis	
Esotropia	65 (82)
Infantile	51 (65)
Acquired	12 (15)
Sensory	2 (3)
Exotropia	12 (15)
Acquired	6 (8)
Primary	3 (4)
Sensory	3 (4)
Hypertropia	1 (1)
Unclear	1 (1)
Presenting diagnosis	
Esotropia	27 (34)
Acquired	2 (3)
Consecutive	3 (4)
Infantile	1 (1)
Recurrent	6 (8)
Residual	2 (3)
Sensory	2 (3)
Unspecified	11 (14)
Exotropia	47 (59)
Consecutive	28 (35)
Recurrent	4 (5)
Sensory	6 (8)
Unspecified	9 (11)
Hypertropia	5 (6)

Median preoperative alignment at distance was 25^{Δ} (range, 6^{Δ}-80^{Δ}) for esotropia, 30^{Δ} (range, 8^{Δ}-95^{Δ}) for exotropia, and 12^{Δ} (range, 3^{Δ}-30^{Δ}) for vertical deviation. Postoperative motor alignment outcomes are presented in Table 2.

Frequency of Diplopia after Strabismus Surgery

At 6 weeks, 63 of 78 patients (81%) reported no diplopia in any gaze and 15 (19%) reported some diplopia in at least 1 direction of gaze (Table 3), of whom 13 (17%) reported diplopia in straight-ahead distance gaze and 8 (10%) reported diplopia in reading gaze (frequencies in Table 3). Seven of the 8 with diplopia in reading gaze also had diplopia in straight-ahead distance gaze. One patient (1%; 95% CI, 0%-7%) had constant diplopia in straight-ahead distance and constant diplopia in reading gaze. No other patient had constant diplopia for straight-ahead distance or reading gaze. Data were missing for 1 patient at the 6-week follow-up.

At 1 year, 43 of 51 patients (84%) reported no diplopia in any gaze and 8 (16%) reported some diplopia in at least 1 direction of gaze (Table 3), of whom 7 (14%) reported diplopia in straight-ahead distance gaze and 4 (8%)

Table 2. Postoperative motor alignment outcomes at 6 weeks and 1 year after strabismus surgery in adults with childhood-onset strabismus

Preoperative status	Outcome alignment, PD[a]	
	6-weeks	1 year
Esotropia	3.5 ET (14 XT to 20 ET)	4 ET (6 XT to 25 ET)
Exotropia	4 ET (16 XT to 30 ET)	3 ET (10 XT to 25 ET)
Vertical	4 HT (2 to 6 HT)	2.5 HT (1 to 10 HT)

ET, esotropia; *HT*, hypertropia; *PD*, prism diopter; *XT*, exotropia.
[a]Medians and range, measured by prism and alternate cover test at distance fixation.

Table 3. Diplopia frequencies at 6 weeks and 1 year after strabismus surgery in adults with childhood-onset strabismus

Frequency	6 weeks (n = 78), no. (%)	1 year (n = 51), no. (%)
No diplopia in any gaze	63 (81)	43 (84)
Diplopia in any gaze	15 (19)	8 (16)
Diplopia in straight ahead distance gaze	13 (17)	7 (14)
Rarely	4 (5)	1 (2)
Sometimes	4 (5)	4 (8)
Often	4 (5)	1 (2)
Always	1 (1)	1 (2)
Diplopia in reading gaze	8 (10)	4 (8)
Rarely	4 (5)	1 (2)
Sometimes	2 (3)	0 (0)
Often	1 (1)	2 (4)
Always	1 (1)	1 (2)

reported diplopia in reading gaze (frequencies in Table 3). All 4 patients who had diplopia in reading gaze also had diplopia for straight-ahead distance gaze. One patient (2%; 95% CI, 0%-10%) had constant diplopia in straight-ahead distance and also in reading gaze with consecutive esodeviation (the same patient who had constant diplopia at 6 weeks). No other patient had constant diplopia for straight-ahead distance or reading gaze. The diplopia could be relieved using base out prism and was therefore not truly "intractable." Twenty-eight patients were lost to follow-up at 1 year.

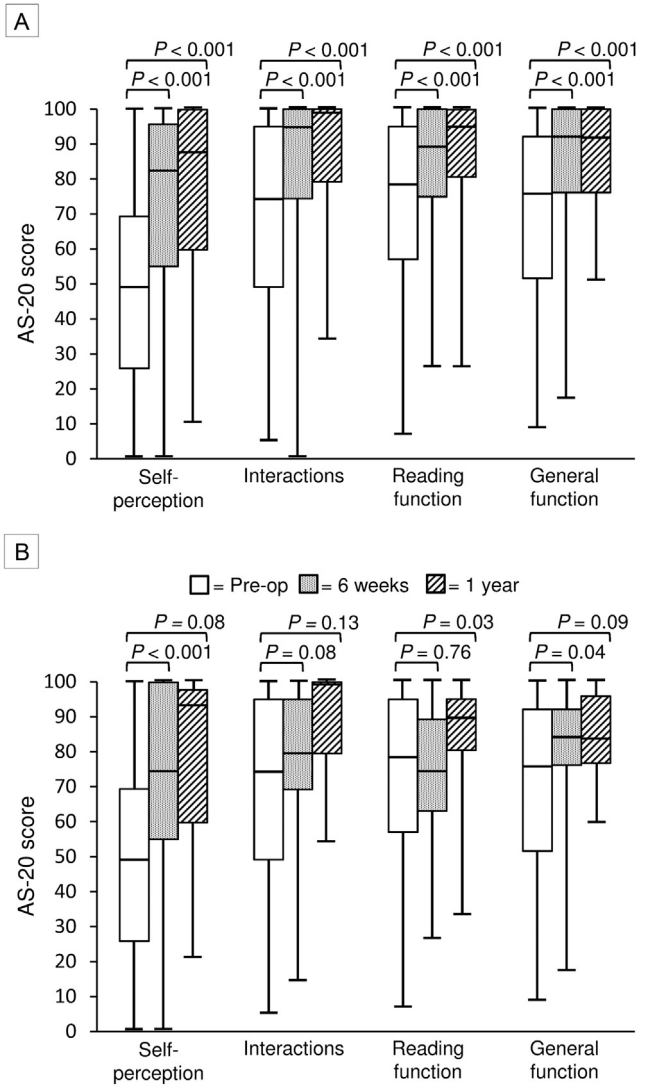

FIG 1. Adult Strabismus-20 health-related quality of life scores; preoperatively, 6 weeks and 1 year postoperatively in adults undergoing surgery for childhood-onset strabismus, both (A) for patients overall and (B) for patients with postoperative diplopia. Boxes represent 1st quartile, median, and 3rd quartile values; whiskers represent extreme values. All domain scores improved significantly from pre- to postoperatively overall at both 6 weeks and 1 year ($P < 0.001$ for all comparisons). For patients with diplopia postoperatively, scores also improved, although significance was limited by the small sample size.

Health-related Quality of Life (HRQOL) Outcomes

Across all patients, AS-20 scores significantly improved on all 4 domains at both 6 weeks and 1 year (Figure 1). At 6 weeks, self-perception improved by a mean of 25 points (95% CI, 19-31; $P < 0.001$); interactions, by 14 points (95% CI, 9-18; $P < 0.001$); reading function, by 10 points (95% CI, 5-15; $P < 0.001$); and general function, by 13 points (95% CI, 8-18; $P < 0.001$). See Figure 1. At 1 year, self-perception improved by 32 points (95% CI, 23-40; $P < 0.001$); interactions, by 19 points (95% CI, 12-26; $P < 0.001$); reading function, by 14 points (95% CI, 9-19; $P < 0.001$); and general function, by 15 points (95% CI, 11-20; $P < 0.001$). See Figure 1.

Regarding the subset of 15 patients with new-onset diplopia, at 6 weeks, AS-20 scores also improved, with a mean change of 23 points (95% CI, 11-34; $P < 0.001$) for self-perception, 10 points (95% CI, 0-19; $P = 0.08$) for interactions, 1 point (95% CI, −11 to 12; $P = 0.76$) for reading function, and 6 points (95% CI, −1 to 13; $P = 0.04$) for general function (Figure 1). For the 8 patients with diplopia at 1 year, scores also improved in all domains: self-perception, by 21 points (95% CI, −2 to 44; $P = 0.08$);

interactions, by 13 points (95% CI, −3 to 29; $P = 0.13$); reading function, by 16 points (95% CI, 2-30; $P = 0.03$); and general function, by 11 points (95% CI, 0-22; $P = 0.09$). See Figure 1.

Discussion

In this study evaluating postoperative diplopia after strabismus surgery in adults with previously nondiplopic childhood strabismus, the rate of constant diplopia was low (2% at 1 year). Considering any diplopia in any gaze position, we found the rate of postoperative diplopia to be 19% at 6 weeks and 16% at 1 year. Considering only straight-ahead distance gaze (any frequency), the rate was 17% at 6 weeks and 14% at 1 year.

Previous studies have evaluated the rate of new-onset diplopia following surgery for nondiplopic strabismus. Kushner[4] described a large series of 424 nondiplopic adults undergoing strabismus surgery and found 40 (9%) experienced temporary postoperative diplopia that resolved by 6 weeks, and 3 (0.8%) had persistent diplopia at 6 weeks. Patients with only "momentary diplopia lasting several seconds- with shift of gaze direction, with fatigue, or upon first arising" were considered nondiplopic. The 3 patients in Kushner's study[4] with persistent diplopia at 6 weeks were described as having intractable diplopia that could not be suppressed at any angle of under- or overcorrection and could not be fused with prism. Scott and colleagues[6] reported postoperative diplopia in 16 patients (4%) in a series of 367 patients without preoperative diplopia. Of these, 5 (1%) continued to have diplopia postoperatively, but it was not stated whether or not diplopia was considered "intractable." Gill and Drummond[5] reported postoperative diplopia in 6 of 137 (4%) visually mature patients without preoperative diplopia: 4 (3%) of these patients had continued diplopia, but it is unclear whether or not diplopia was considered "intractable." In the present study, we found a comparable rate of diplopia when considering only constant diplopia in straight-ahead and reading gaze (2%), but a higher rate of diplopia when considering any frequency in any gaze position.

The 1 patient with constant diplopia straight-ahead and reading was a 33-year-old man with a history of infantile esotropia and a large-angle exotropia preoperatively due to lost medial rectus muscle after previous esotropia surgery as a young child. After his most recent surgery, he had a small-angle esotropic alignment with "always" horizontal diplopia in all directions at 6 weeks, which did not improve at 1 year. Nevertheless, this diplopia was not truly intractable because he was able to suppress with 25^{Δ} of base-out prism in the office (recreating an exodeviation). He was unable to tolerate prism glasses (because they "made him sick") but reported being very happy with the improvement in alignment and not bothered by his diplopia, stating he knew "which is the real image." He was not interested in further surgery or in using a blurring lens or occlusion to reduce diplopia.

In the present study, we used a prospective standardized questionnaire to evaluate diplopia in contrast to retrospective review of the medical record. This standardized rating of diplopia frequency using a patient-reported outcome measure may have resulted in more frequent reporting of diplopia than previous studies. Regarding inconsistencies between patient self-report and documentation in the medical record, Valikodath and colleagues[8] reported symptoms being more frequently recorded using a questionnaire. In addition, our diplopia questionnaire[7] allows reporting of diplopia in all gaze positions, whereas previous studies either evaluated only straight-ahead distance gaze[6] or did not specify which gaze positions were evaluated.[4,5] Regarding diplopia frequency, our diplopia questionnaire[7] provides a standardized rating scale enabling a 5-level gradation of diplopia severity, whereas in previous studies, the rating of frequency was less standardized.

We are not aware of any prior studies evaluating HRQOL outcomes of patients with new postoperative diplopia. We have previously reported[3] improved function-related HRQOL after surgery for childhood-onset strabismus in a subset of this overall cohort, but we have not specifically reported HRQOL in patients with postoperative diplopia. In the present study, we found that in previously nondiplopic patients with new postoperative diplopia, there was improvement in all four AS-20 domains based on mean scores, although not all of the improvements were statistically significant, likely due to the small sample size. Of the four domains, reading function was least improved at 6 weeks in diplopic patients (mean, 1 point) but had improved by a mean of 16 points at 1 year. Some patients reported that the new diplopia was not bothersome, ignoring the second image. One patient with "often" diplopia stated, "Even knowing what I know now, I would still have done it." Such examples illustrate that even when diplopia occurs, improved quality-of-life from better alignment may outweigh the disadvantages of new diplopia.

There are several limitations to this study. We did not have complete follow-up data, because a proportion of patients did not return for follow-up at 1 year (79 patients were initially enrolled, and 28 [35%] patients did not have 1-year data). Therefore, diplopia at 1 year may be overreported as a proportion because patients with symptoms may have been more inclined to follow-up. The present study is also limited by a relatively small sample size. We did not evaluate change in AS-20 score in a control group, but a previous study documented marked differences between successfully and unsuccessfully treated patients undergoing strabismus surgery[1]; therefore, it is unlikely that improvements we found in the present study represent a placebo effect.

In adults with previously nondiplopic childhood-onset strabismus, we found the rate of constant diplopia to be low and comparable with that reported in previous studies. Nevertheless, intermittent diplopia, when considering any gaze position, occurs more frequently. Even when

postoperative diplopia occurs, HRQOL often markedly improves. These data are helpful for counseling nondiplopic patients regarding the risks and benefits of strabismus surgery.

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