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Rates of Reoperation in Duane Retraction Syndrome

Han Woong Lim, MD, PhD,^{1,2} Bryce Hwang, MD,^{1,3} Cyril Archambault, MD,⁴ Scott R. Lambert, MD¹

Purpose: To investigate the types of strabismus surgeries performed and the reoperation rate in patients with Duane retraction syndrome (DRS).

Design: Retrospective cohort analysis.

Participants: An insurance claims data set was used to identify patients diagnosed with DRS between 2007 and 2021.

Methods: We recorded the type of strabismus surgery performed and the timing and frequency of reoperations. The hazard ratios (HRs) for reoperation were estimated according to the surgical methods using Cox regression analysis.

Results: Of the 9435 patients diagnosed with DRS, 1023 (10.8%) underwent ≥ 1 strabismus operation. The median age at surgery was 5.0 years, and patients were followed for an average of 3.8 ± 3.0 years after their initial strabismus surgery. Most of the surgeries only involved horizontal muscle(s) ($n = 734$ [71.7%]). However, some patients underwent surgeries on vertical muscle(s) ($n = 132$ [12.9%]), vertical muscle(s) with transposition ($n = 102$ [10.0%]), and horizontal muscle with transposition ($n = 51$ [5.0%]). The estimated 5-year rate of reoperation was 18.2% (95% confidence interval [CI], 15.0%–22.2%). Compared with surgery on horizontal muscle(s) only, vertical muscle surgery (HR, 2.01; 95% CI, 1.30–3.11; $P = 0.002$) and vertical muscle surgery coupled with transposition (HR, 1.79; 95% CI, 1.06–3.02; $P = 0.03$) had an increased risk of reoperation.

Conclusions: Strabismus surgery on ≥ 1 horizontal muscles is the most common type of strabismus surgery performed on patients with DRS. Approximately 1 in 7 patients with DRS who had strabismus surgery underwent a reoperation. Patients who underwent vertical muscle surgery had a higher risk of undergoing a reoperation.

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Duane retraction syndrome (DRS) is the most common type of congenital aberrant ocular innervation.¹ It is characterized by globe retraction and palpebral fissure narrowing on adduction, with limitation of abduction, adduction, or both.² The incidence of DRS is $\sim 1\%$ to $\sim 4\%$ of all strabismus cases.³ Duane retraction syndrome is caused by congenital hypoplasia or absence of the sixth nerve and nucleus with aberrant innervation by branches of the oculomotor nerve resulting in cocontraction of the medial and lateral rectus muscles on adduction.^{4,5}

There are various options for the surgical treatment of DRS. However, the surgical treatment of DRS is challenging because each patient has a unique amount of innervation of the lateral rectus muscle, as well as muscle contractures, leading to a diversity of clinical presentations.^{6–9} A few previous studies and case reports have examined the rate of reoperation after strabismus surgery in patients with DRS. However, these studies are limited by small sample sizes and single-center design.^{10–14} The reoperation rate in a large cohort of patients with DRS has not been previously investigated.

We conducted this study to investigate rates of reoperation and to characterize risk factors associated with

reoperations in patients with DRS using a commercial insurance claims data set.

Methods

Data Collection

This population-based retrospective cohort study was performed using claims data from the IBM MarketScan Research Database. This database comprises deidentified commercial claims from a large claims data warehouse from 2007 to 2022, covering a total of approximately 164 million lives from a geographically diverse population spanning all 50 United States states. This database provides demographic, socioeconomic, and medical claims data for inpatient and outpatient services, including surgical services. The medical claims data use the International Classification of Diseases (ICD) 9th (ICD-9-Clinical Modification) and 10th (ICD-10-Clinical Modification) Revisions diagnosis codes and Current Procedural Terminology (CPT) version 4 procedure codes. This analysis was deemed exempt from Stanford University Institutional Review Board approval. Per Population Health Sciences Data Core policy to prevent deidentification, all cell sizes < 16 are reported as < 16 .

Table 1. Demographics of Patients with Duane Retraction Syndrome

	No Surgery (n = 8412)	Surgery (n = 1023)	Total (n = 9435)	P Value
Yr of diagnosis				< 0.001
2007–2009	2256	313	2569	
2010–2012	2570	342	2912	
2013–2015	1651	186	1837	
2016–2018	1248	128	1376	
2019–2021	687	54	741	
Age (yrs), median (IQR)	11 (4–32)	5 (1–19)	11 (3–31)	< 0.001
Sex				0.44
Male	3688	462	4150	
Female	4724	561	5285	
Yrs of continuous enrollment	4.4 ± 3.2	4.9 ± 3.4	4.4 ± 3.2	< 0.001

IQR = interquartile range.

Data Extraction and Analysis

All beneficiaries with newly diagnosed DRS who had undergone strabismus surgery were included if they had continuous enrollment from 6 months before the first diagnosis of DRS and 6 months after the initial strabismus surgery. The first diagnosis of DRS was defined as the first date the ICD-9 code 378.71 or the ICD-10 code H50.81x was used for a patient encounter. Data extracted for each patient included age at diagnosis of DRS, age at first strabismus surgery, gender, period of continuous enrollment, CPT codes for initial strabismus operation performed, and dates of strabismus surgery.

The CPT codes used for the first strabismus surgery for each patient with DRS were analyzed: horizontal muscles only (CPT codes 67311 or 67312), vertical muscles only (CPT codes 67314, 67316, 67318), or both horizontal and vertical muscles (both horizontal muscle and vertical muscle CPT code), and chemodeneration of extraocular muscles (CPT 67345). The use of transposition of the muscles during the operation was determined using the add-on CPT code 67320. A strabismus surgery performed on a different date after the initial operation was considered a reoperation. Use of adjustable sutures was determined using CPT code 67335. Because the reoperation code (CPT 67332) for strabismus can be used for patients who have restriction of the

extraocular muscles, we did not use this code for determining if a reoperation had been performed.

Comparisons between patients who did and did not undergo reoperations were made using the chi-square test for age, gender, surgical methods, reoperation rate, and a 1-way analysis of variance for continuous variables with ≥ 3 groups. This was followed by a secondary post hoc analysis for surgical methods. The Mann–Whitney *U* test was used to compare age at the time of the initial surgery, the follow-up period after the initial surgery, and the continuous enrollment period after the initial surgery between the 2 groups. Cox proportional hazards regression analyses were performed to evaluate the effects of sex, age, and surgical methods at the initial surgery on reoperation. Hazard ratios (HRs) were calculated along with corresponding confidence intervals (CIs). A *P* value of < 0.05 was considered statistically significant. All statistical analyses were conducted using R software (version 4.1.1).

Results

Demographics

A total of 11 252 719 663 claims for 148 147 615 patients were analyzed in the IBM MarketScan Database

Table 2. Comparison of Patients Undergoing a Single Strabismus Surgery vs. ≥ 2 Strabismus Surgeries

	Single Surgery (n = 883)	≥ 2 Surgeries (n = 140)	P Value
Yr of first operation			0.04
2007–2009	262	51	
2010–2012	297	45	
2013–2015	161	25	
2016–2018	114	< 16	
2019–2021	49	< 16	
Age (yrs), median (IQR)	6.0 (2.0–19.0)	4.0 (2.0–20.0)	0.82
Sex			0.60
Male	404	58	
Female	479	82	
Yrs of continuous enrollment	4.7 ± 3.4	5.7 ± 3.7	< 0.001
Yrs of follow-up after initial surgery	3.7 ± 3.0	4.6 ± 3.4	< 0.001
Yrs to reoperation	NA	1.5 ± 1.7	
Adjustable suture use	103	< 16	0.35

IQR = interquartile range; NA = not applicable.

Table 3. Comparison of Initial Surgery Type between Single Operation and Reoperation Groups

Type of Operation	Single Surgery	≥ 2 Surgeries
Chemodeneration	< 16	< 16
Horizontal muscle(s) without transposition	646	88
Horizontal muscles(s) with transposition	45	< 16
Procedure involving vertical muscle(s) without transposition	103	29
Procedure involving vertical muscle(s) with transposition	85	17

Percentages and totals are not given to preclude calculation of cells < 16.

(version 3.0) and MarketScan Medicare Database (version 3.0) for the period 2007-2022, from which 9435 patients were diagnosed with DRS and met inclusion criteria. The median age at the first date of diagnosis was 11 (interquartile range, 3–31) years, with a female predominance (5285/9435 [56.0%]). The mean number of years of continuous enrollment was 4.4 ± 3.2 years (Table 1).

Baseline Characteristics

Strabismus surgery was performed on 1023 of 9435 patients (10.8%) diagnosed with DRS between 2007 and 2022 who also met the enrollment exclusion criteria. Of the 1023 patients who underwent strabismus surgery, 140 (13.7%) underwent a reoperation. The median age at first strabismus surgery was 4.0 years (interquartile range, 2.0–20.0) for patients who underwent ≥ 2 strabismus surgeries and 6.0 years (interquartile range, 2.0–19.0) for patients that underwent a single strabismus surgery ($P = 0.82$). There was a predominance of females in both the single and multiple-surgery groups (54.2% vs. 58.6%), but that there was no difference between the percentage of females in the single-surgery group and the multiple-surgery group ($P = 0.60$). The total years of follow-up were greater for patients who underwent reoperation (5.7 ± 3.7 years) compared with patients who had a single operation (4.7 ± 3.4 years; $P < 0.001$). The mean number of years of follow-up after initial surgery was less than in the single-operation group compared with the multiple-operation group (3.7 ± 3.0 vs. 4.6 ± 3.4 years, $P < 0.001$, Table 2).

Characteristics of Initial Surgeries

The majority of initial surgeries were performed on horizontal muscle(s) (734/1023 [71.8%]). Less commonly, initial surgeries were performed involving vertical muscle(s) without transposition (132/1023 [12.9%]), vertical muscle(s) with transposition (102/1023 [10.0%]), horizontal muscles with transposition (51/1023 [5.0%]), and chemodeneration (< 16/1023). The 5-year estimate for the risk of reoperation was 18.2% (95% CI, 15.0%–22.2%). The rate of reoperation was highest among patients whose initial

Table 4. Hazard Ratios for Multivariate Cox Regression by Prognostic Factor for Reoperation Rate

	Hazard Ratios (95% CI)	P Value
Age	1.00 (0.99–1.01)	0.97
Sex		
Male	1.22 (0.86–1.73)	0.27
Female	Reference	
Adjustable suture use		
Use	Reference	
Nonuse	0.79 (0.43–1.45)	0.44
Initial surgical measure		
Horizontal muscle(s) without transposition	Reference	
Horizontal muscle(s) with transposition	1.21 (0.53–2.78)	0.65
Procedure involving vertical muscle(s) without transposition	2.01 (1.30–3.11)	0.002
Procedure involving vertical muscle(s) with transposition	1.79 (1.06–3.02)	0.029

CI = confidence interval.

surgery involved vertical muscle(s) with transposition (29/132 [22.0%]), followed by vertical muscle(s) without transposition (17/102 [16.7%]) and horizontal muscle surgery (88/734 [12.0%]) (Fig 1). For horizontal muscle surgery with transposition, the number of reoperations was < 16 (Table 3). Reoperations were performed more often on patients who underwent procedures involving vertical muscle(s) without transposition compared with other surgeries (29/132 [22.0%] vs. 111/891[12.5%], $P = 0.005$). Adjustable sutures were performed in 115/1023 (11.2%) of patients. The use of adjustable sutures was not different among those that did and did not undergo a reoperation (12/115 [10.4%] vs. 128/908 [14.1%], $P = 0.35$).

Risk Factors for Undergoing a Reoperation

Cox regression analysis found that surgeries that involved vertical muscle(s) without transposition (HR, 2.01; 95% CI, 1.30–3.11; $P = 0.002$), as well as surgeries that involved vertical muscle(s) with transposition (HR, 1.79; 95% CI, 1.06–3.02; $P = 0.03$) were associated with a higher rate of reoperation. Compared with patients who did not receive a reoperation, there were no differences in age, gender, or adjustable suture rates (Table 4).

Discussion

In this claims-based analysis, approximately 11% of patients with DRS underwent strabismus surgery. Approximately 70% of patients with DRS who underwent strabismus surgery had surgery on horizontal muscle(s) only, and the reoperation rate for this group was 12.0%. The risk of reoperation differed according to the type of initial operation. Surgeries on vertical muscles, with, or without transposition, were associated with an increased

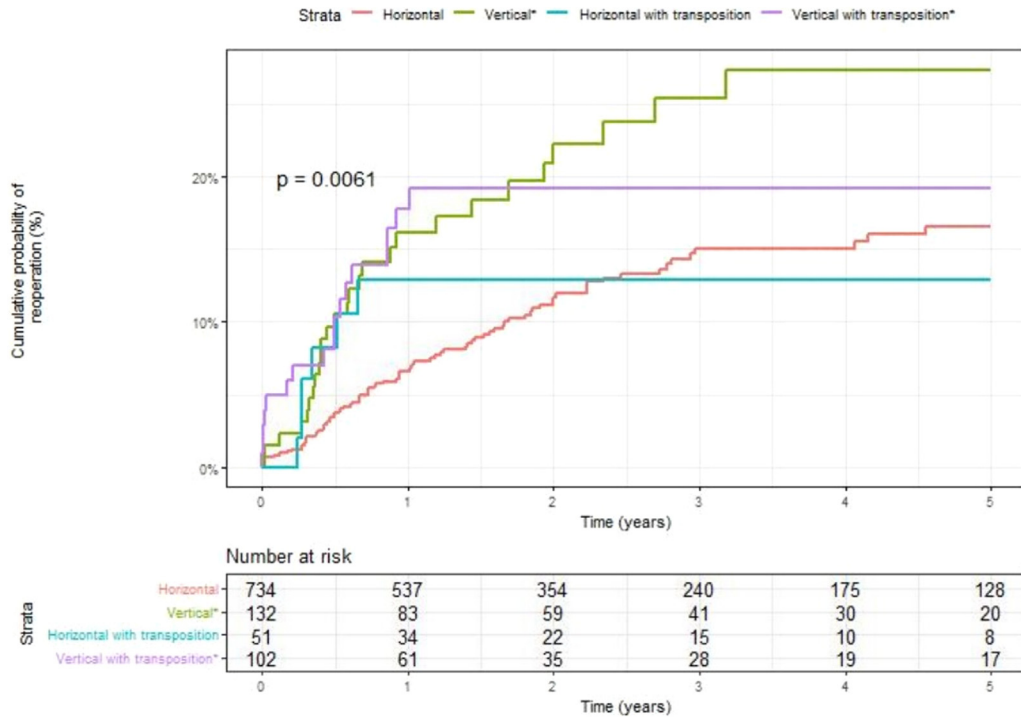


Figure 1. Kaplan–Meier plot of the cumulative probability for reoperation after different types of strabismus surgery for patients with Duane retraction syndrome. The number of patients at risk over time is shown in the table below the plot.

HR for reoperation. The estimated 5-year reoperation rate was 18.2%. Age did not affect the reoperation rate. The risk of reoperation was approximately 2 times higher for procedures involving vertical muscle(s) with transposition (22%) compared with only horizontal muscle surgery (12%).

It is likely that our analysis was an underestimate of the true incidence of strabismus surgery in patients with DRS since our cohort of patients was only followed on average for 4.3 years. The incidence of strabismus surgery was lower in the 2019 to 2021 time period (8%), perhaps because these patients weren't followed as long as patients in other time periods or because many elective surgeries were postponed during the coronavirus disease 2019 pandemic.

Previous studies of strabismus surgery in patients with DRS have reported success rates ranging from 50% to 94%, but were limited by variable definitions of success, small sample sizes, and a short postoperative follow-up.^{10–14} Barbe et al¹⁰ reported postoperative ocular alignment of ≤ 15 prism diopters (PDs) and ≤ 5 PD in 93% and 66% of the patients in their series, respectively. In a clinic-based study of 65 patients, Chua et al¹¹ reported postoperative alignment of ≤ 15 PD in 86% of their cases and ≤ 5 PD in 50%. More recently, Sheath et al¹⁴ reported that motor alignment in primary position, resolution of an abnormal head position, and correction of overshoots were achieved in 74%, 81%, and 71% of patients, respectively. Furthermore, they reported a 19% (14 among 73 patients with DRS who underwent strabismus surgery)

reoperation rate. We found a 13.7% overall reoperation rate in a large cohort of patients with DRS using commercial insurance claims, which is comparable to these studies. The advantages of our study over previous reports were a longer follow-up interval, a larger sample size, and nationally representative data.

Most patients with DRS in our data set only underwent horizontal muscle(s) surgery. Although CPT codes do not allow us to determine whether a recession or resection procedure was performed, it is likely that most procedures were recessions since horizontal muscle resections in patients with DRS may impair ductions and worsen globe retraction.¹⁵ Recession of a horizontal rectus muscle is commonly used to improve an abnormal head position and ocular alignment in primary position in patients with DRS.^{10,13} In a study of 59 patients with DRS, surgical success was reported in 93% of patients after a horizontal muscle recession.¹⁰ Similarly, our results show that horizontal muscle(s) surgery was the most commonly performed type of strabismus surgery performed for DRS and was associated with the lowest reoperation rate (12.0%).

Vertical muscle transposition procedures have been reported to be more effective in improving abduction and expanding the field of a binocular single vision.^{16–19} On the other hand, transposition procedures increase the risk of inducing vertical deviations and anterior segment ischemia, especially if combined with horizontal rectus surgery.^{20–24} Foster²⁵ has warned that ipsilateral medial rectus recession should be avoided in patients undergoing a vertical transposition procedure

because of the risk of decreased adduction and late over-corrections. In our series, reoperation rates and HRs of reoperation were higher in patients who underwent horizontal and vertical muscle surgery coupled with transposition compared with other strabismus surgeries.

Y splitting of the lateral rectus muscle has been found to be an effective surgical procedure to reduce upshoots and downshoots in patients with DRS and a tight lateral rectus muscle.^{26,27} These patients typically have an exotropia in primary position and limited adduction in the involved eye. Y splitting is often coded as a horizontal muscle and transposition procedure. This group of patients presumably accounts for most of the claims billed as horizontal muscle surgery coupled with transposition. However, it is also possible that this code was used when offsetting horizontal muscles in patients with an A or V pattern. This group of patients had a reoperation rate of 11.2%.

This study has several limitations. First, it was conducted using insurance claims data, which do not provide information on the angle of deviation, amount of gaze limitation, presence of abnormal head posture, fixation preference, overshoot or undershoot, visual acuity, stereoacuity, or surgical dosing data. Furthermore, it was not possible to distinguish the type and laterality of DRS. Second, miscoding may have occurred if a provider submitted the wrong diagnosis or procedure code. Third, we

could not investigate data for patients outside their enrollment period; hence, we may have missed patients who were diagnosed and underwent surgery for DRS before enrollment. Overall, this limits the ability of this study to draw conclusions about the effectiveness of specific surgical approaches. However, this claims-based study does suggest hypotheses worth further investigating with patient-level data.

In conclusion, our results using medical claims data that reflects the real-world practice of DRS treatment in the United States showed that strabismus surgery was performed only on the horizontal muscles in about 70% of cases and the overall 5-year risk of reoperation was 18.2%. The initial surgery type was the strongest independent predictor of undergoing a reoperation and the reoperation rate was higher for patients who underwent surgery involving vertical muscles.

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No animal subjects were used in this study.

Author Contributions:

Conception and design: Lim, Hwang, Archambault, Lambert

Data collection: Lim, Hwang, Lambert

Analysis and interpretation: Lim, Hwang, Archambault, Lambert

Obtained funding: Lim, Lambert

Overall responsibility: Lim, Hwang, Archambault, Lambert

Abbreviations and Acronyms:

CI = confidence interval; **CPT** = Current Procedural Terminology; **DRS** = Duane retraction syndrome; **HR** = hazard ratio; **ICD** = International Classification of Diseases; **PD** = prism diopters.

Keywords:

Duane retraction syndrome, Reoperation, Strabismus surgery, Y splitting, Insurance claims data.

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