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## Characterization of Errors in Retinopathy of Prematurity Diagnosis by Ophthalmologists-in-Training in the United States and Canada

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# Characterization of Errors in Retinopathy of Prematurity Diagnosis by Ophthalmologists-in-Training in the United States and Canada

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## ABSTRACT

**Purpose:** To identify the prominent factors that lead to misdiagnosis of retinopathy of prematurity (ROP) by ophthalmologists-in-training in the United States and Canada.

**Methods:** This prospective cohort study included 32 ophthalmologists-in-training at six ophthalmology training programs in the United States and Canada. Twenty web-based cases of ROP using wide-field retinal images were presented, and ophthalmologists-in-training were asked to diagnose plus disease, zone, stage, and category for each eye. Responses were compared to a consensus reference standard diagnosis for accuracy, which was established by combining the clinical diagnosis and the image-based diagnosis by multiple experts. The types of diagnostic errors that occurred were analyzed with descriptive and chi-squared analysis. Main outcome measures were frequency of types (category, zone, stage, plus disease) of diagnostic errors; association of errors in zone, stage, and plus disease diagnosis with incorrectly identified category; and performance of ophthalmologists-in-training across postgraduate years.

**Results:** Category of ROP was misdiagnosed at a rate of 48%. Errors in classification of plus disease were most commonly associated with misdiagnosis of treatment-requiring (plus error rate = 16% when treatment-requiring was correctly diagnosed vs 81% when underdiagnosed as type 2 or pre-plus; mean difference: 64.3; 95% CI: 51.9 to 76.7;  $P < .001$ ) and type 2 or pre-plus (plus error rate = 35% when type 2 or pre-plus was correctly diagnosed vs 76% when overdiagnosed as treatment-requiring; mean difference: 41.0; 95% CI: 28.4 to 53.5;  $P < .001$ ) disease. The diagnostic error rate of postgraduate year (PGY)-2 trainees was sig-

nificantly higher than PGY-3 trainees (PGY-2 category error rate = 61% vs PGY-3 = 35%; mean difference, 25.4; 95% CI: 17.7 to 33.0;  $P < .001$ ).

**Conclusions:** Ophthalmologists-in-training in the United States and Canada misdiagnosed ROP nearly half of the time, with incorrect identification of plus disease as a leading cause. Integration of structured learning for ROP in residency education may improve diagnostic competency.

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## INTRODUCTION

Retinopathy of prematurity (ROP) is a vasoproliferative disease that affects the retinas of premature infants. This disease continues to be a common cause of childhood vision loss in the United States and worldwide.<sup>1,2</sup> The Early Treatment for ROP and the Cryotherapy for ROP study findings have paved the way for screening practices and interventions; however, diagnosis and management of ROP continue to be imperfect due to inadequate exposure to ROP during residency and fellowship training.<sup>1,3,4</sup> As a result, a small number of skilled ophthalmologists are capable and willing to provide ROP services.<sup>5</sup>

In a survey of third-year ophthalmology residents and program directors, 66% of board-eligible ophthalmologists reported conducting fewer than 20 ROP examinations during training, and 19% reported that residents do not perform any examinations.<sup>5</sup> Due to the overall minimal number of opportunities to examine patients for ROP during

residency, only 17% indicated feeling confident on completion of training.<sup>5</sup> Moreover, 4% of residents indicated that structured evaluations were used.<sup>5</sup> Although fellowship programs at institutions may divert ROP cases away from residents, retina and pediatric ophthalmology trainees may not typically achieve full competency in ROP care.<sup>6-8</sup>

Currently, there is little standardization in ROP education, which varies the training experience.<sup>9</sup> Among retina fellows, a study found that type 2 ROP was misdiagnosed as treatment-requiring ROP 47% of the time.<sup>8</sup> Similarly, in a study of pediatric ophthalmology fellows, trainees diagnosed type 2 and treatment-requiring ROP with 50% sensitivity.<sup>7</sup>

Given that ROP screening may be conducted by comprehensive ophthalmologists, it is critical that ophthalmologists-in-training develop competency in ROP management. However, little data exist on the evaluation of common errors in ROP diagnosis in ophthalmology training programs. The purpose of this study was to characterize common errors in

ROP diagnosis by ophthalmologists-in-training in the United States and Canada.

## METHODS

The Weill Cornell Medical College Human Studies Committee prospectively approved all aspects of the use and analysis of retinal images and educational material used in this study. Administration of the web-based system was also reviewed by The Weill Cornell Medical College Human Studies Committee. This was considered to be research in an established or commonly accepted educational setting involving normal educational practices, such as research on the effectiveness of instructional techniques, curricula, and instructional strategies. The research adhered to the tenets of the Declaration of Helsinki.

### Case Acquisition

The web-based system used for ROP assessment has been previously fully described.<sup>1</sup> The program created included 16 unique cases consisting of wide-

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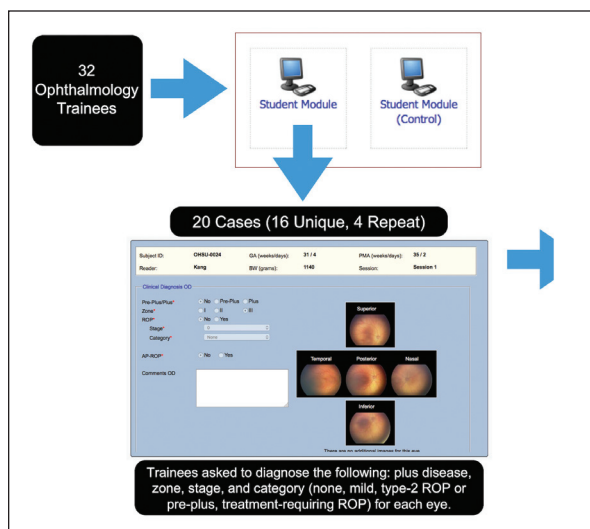
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**Figure 1.** Design of study methods for the assessment of diagnostic competency in retinopathy of prematurity (ROP). Thirty-two ophthalmologists-in-training were directed to a web-based module consisting of 20 cases of ROP. Trainees selected the diagnosis for zone (I, II, III), stage (1-5), category (none, mild, type 2 or pre-plus, and treatment-requiring), and plus disease (none, pre-plus, plus) for each eye presented. The rate at which trainees accurately diagnosed ROP was determined based on a consensus reference standard diagnosis.

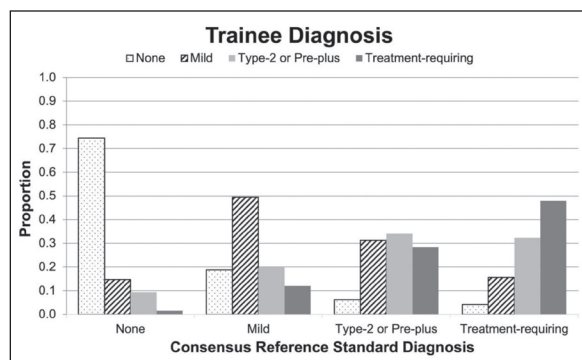
field fundus images selected from a bank of 2,500 web-based cases. Four of these cases were presented twice to the participants for a total of 20 cases.

### Study Participants and Study Design

Ophthalmologists-in-training from five training programs in the United States and one program in Canada were invited to participate in the study.<sup>1</sup> The trainees accessed the secure web-based platform, where they completed the assessment composed of the 20 cases selected (**Figure 1**). In each case, the trainees diagnosed ROP based on imaging of 2 eyes. In total, trainees evaluated 32 unique eyes for ROP and determined the following criteria based on the international standardized classification system: plus disease (none, pre-plus, plus), zone (I, II, III), stage (1, 2, 3, 4, 5), category (none, mild, type 2 or pre-plus, treatment requiring), and aggressive posterior ROP (yes, no). On completion of the cases, recorded answers could not be changed. The 20 cases selected varied in the category of disease; however, the same cases were presented to all participants, regardless of year of training.

### Consensus Reference Standard Diagnosis

The methods for deriving the consensus reference standard diagnosis for each case in this study



**Figure 2.** Distribution of ophthalmologists-in-training's responses when diagnosing category of retinopathy of prematurity. The correct category of disease (none, mild, type 2 or pre-plus, and treatment-requiring) is based on a consensus reference standard diagnosis.

have been previously explained.<sup>1</sup> To summarize, the reference standard diagnosis is based on the findings identified by the ROP expert on indirect ophthalmoscopy combined with the diagnosis individually determined by three additional ROP experts when examining digital color fundus photographs as an alternative to the gold standard clinical examination.

### Data Analysis

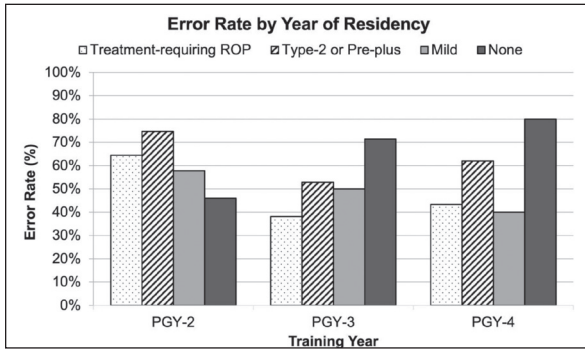
Statistical analysis was conducted using SAS software (SAS Institute), and a two-sided *P* value of less than .05 was deemed to be statistically significant. Responses for each postgraduate year (PGY) were analyzed using univariate relationships without adjustment for other covariates using the chi-squared test or Fisher exact test. The participants' responses were graded based on the consensus reference standard diagnosis.<sup>1</sup>

## RESULTS

A total of 55 ophthalmologists-in-training from five residency programs were directed to the web-based platform. Of these trainees, 46 initiated the assessment and 32 completed the assessment. Performance of the 32 test-takers was analyzed. Stratification of these trainees by PGY was as follows: 15 of 32 trainees (47%) were PGY-2, 7 of 32 trainees (22%) were PGY-3, and 10 of 32 trainees (31%) were PGY-4.

### Diagnostic Error Rate of ROP Category Diagnosis

The overall diagnostic error rate for any category of ROP was 48% (490 of 1,024 responses) across all ophthalmologists-in-training. The most frequently missed diagnoses were for type 2 ROP, with an error rate of 66% (211 of 320 responses), and treatment-requiring ROP, with an error rate of 52% (100 of 192



**Figure 3.** Distribution of errors rates for retinopathy of prematurity (ROP) category diagnosis by training year. Categories of disease include none, mild, type 2 or pre-plus, and treatment-requiring ROP. Postgraduate year (PGY)-2, PGY-3, and PGY-4 ophthalmologists-in-training were included.

responses). For each category of ROP, the distribution of ROP diagnoses, compared to the consensus reference standard diagnosis, are shown in **Figure 2**.

When stratified by PGY, the overall error rate for ROP category diagnosis was as follows: 61% for PGY-2, 35% for PGY-3, and 38% for PGY-4. The diagnostic error rates for each category of ROP by

PGY are shown in **Figure 3**. On average, PGY-2 ophthalmologists-in-training performed significantly worse than PGY-3 trainees (PGY-2 category error rate = 61% vs PGY-3 = 35%; mean difference: 25.4; 95% CI: 17.7 to 33.0;  $P < .001$ ) and significantly worse than PGY-4 trainees (PGY-4 category error rate = 38%; mean difference: 23.1; 95% CI: 16.3 to 30.0;  $P < .001$ ). There was no significant difference in the diagnostic error rate for category between PGY-3 and PGY-4 trainees (mean difference: 2.2; 95% CI: -10.4 to 6.0;  $P = .59$ ).

### Reasons for Incorrect ROP Diagnosis

**Table 1** summarizes the rates at which plus disease, zone, and stage were misdiagnosed in the context of the category being incorrectly selected by the ophthalmologists-in-training. Overall, incorrect classification of plus disease was the factor most commonly associated with misdiagnosis of category (plus error rate = 12% with correct category diagnosis vs 67% when misdiagnosed; mean difference: 55.2; 95% CI: 50.2 to 60.2;  $P < .001$ ). Misdiagnosis of stage (stage error rate

TABLE 1  
**Comparison of Ophthalmologists-in-Training ROP Category Diagnosis to Error Rates of Plus Disease, Zone, and Stage Diagnoses**

Consensus Reference Standard Diagnosis of Category	Resident Diagnosis of Category	Error Rate of Plus Disease Diagnosis, % (SD)	Error Rate of Zone Diagnosis, % (SD)	Error Rate of Stage Diagnosis, % (SD)
Treatment-requiring (n = 192)	Type 2 or pre-plus (n = 62)	80.64 (5.02)	45.16 (6.32)	38.71 (6.19)
	Mild (n = 30)	73.33 (8.07)	63.33 (8.80)	100 (0)
	None (n = 8)	87.50 (11.69)	37.50 (17.12)	87.50 (11.69)
Type 2 or pre-plus (n = 320)	Treatment-requiring (n = 91)	75.82 (4.49)	50.55 (5.24)	49.45 (5.24)
	Mild (n = 100)	82.00 (3.84)	48.00 (5.00)	76.00 (4.27)
	None (n = 20)	95.00 (4.87)	90.00 (6.71)	95.00 (4.87)
Mild (n = 192)	Treatment-requiring (n = 23)	60.87 (10.18)	56.52 (10.34)	86.96 (07.2)
	Type 2 or pre-plus (n = 38)	52.63 (8.10)	26.32 (7.14)	57.89 (8.01)
	None (n = 36)	0.00 (0)	88.89 (5.24)	94.44 (3.82)
None (n = 320)	Treatment-requiring (n = 5)	100 (0)	60.00 (21.91)	100 (0)
	Type 2 or pre-plus (n = 30)	83.3 (6.80)	23.33 (7.72)	100 (0)
	Mild (n = 47)	36.17 (7.01)	59.57 (7.16)	87.23 (4.87)

ROP = retinopathy of prematurity; SD = standard deviation (error rate of plus in the resident diagnosis category)

= 26% with correct category diagnosis vs 72% when misdiagnosed; mean difference: 46.4; 95% CI: 41.0 to 51.8;  $P < .001$ ) was also found to have a significant association with incorrect category diagnosis.

Underdiagnosis of treatment-requiring disease as type 2 or pre-plus was most commonly associated with incorrect identification of plus disease (plus error rate = 16% with correct category diagnosis vs 81% when underdiagnosed as type 2 or pre-plus; mean difference: 64.3; 95% CI: 51.9 to 76.7;  $P < .001$ ).

Overdiagnosis of type 2 or pre-plus disease as treatment-requiring disease was most commonly associated with errors in diagnosis of plus disease (plus error rate = 35% with correct category diagnosis vs 76% when overdiagnosed as treatment-requiring; mean difference: 41.0; 95% CI: 28.4 to 53.5;  $P < .001$ ) and zone (zone error rate = 21% with correct category diagnosis vs 51% when overdiagnosed as treatment-requiring; mean difference: 29.5; 95% CI: 16.6 to 42.3;  $P < .001$ ). Overall, when the category of disease for cases of type 2 or pre-plus ROP was misdiagnosed by trainees, underdiagnosis was the cause 73% of the time (95% CI: 70.0 to 76.2;  $P < .001$ ).

**Figure 3** shows the error rates of ROP category by year of training. PGY-2 ophthalmologists-in-training compared to PGY-3 trainees misdiagnosed stage (PGY-2 stage error rate = 61% vs PGY-3 = 32%; mean difference: 29.1; 95% CI: 21.6 to 36.6;  $P < .001$ ) and plus disease (PGY-2 plus error rate = 51% vs PGY-3 = 30%; mean difference: 20.7; 95% CI: 13.2 to 28.2;  $P < .001$ ) at significantly higher rates. Similarly, error rates of PGY-2 compared to PGY-4 trainees were also significantly different for stage (PGY-4 stage error rate = 39%; mean difference: 22.5; 95% CI: 15.6 to 29.4;  $P < .001$ ) and plus disease (PGY-4 plus error rate = 27%; mean difference: 24.1; 95% CI: 17.5 to 30.7;  $P < .001$ ).

## DISCUSSION

This study examined the error rate at which ophthalmologists-in-training incorrectly categorized ROP and the common factors associated with misdiagnosis. The key findings are: (1) the overall diagnostic error rate for any category of ROP was nearly 50%; (2) incorrectly identifying plus disease was most commonly associated with misdiagnosis of treatment-requiring and type 2 or pre-plus disease; and (3) PGY-2 ophthalmologists-in-training were more likely to misdiagnose ROP compared to PGY-3 and PGY-4 ophthalmologists-in-training.

The first key finding is that ophthalmologists-in-training misdiagnosed all categories of ROP, with underdiagnosis of type 2 ROP as one of the most common errors. Patients defined as having type 2 ROP must be routinely examined for complete vascular development.<sup>8</sup> Progression to treatment-requiring ROP would then require immediate intervention, such as with laser photocoagulation or anti-vascular endothelial growth factor treatment.<sup>8</sup> However, if type 2 ROP is underdiagnosed by ophthalmologists-in-training, there could be a potential delay in the diagnosis of patients who progress from type 2 ROP to treatment-requiring ROP, increasing the risk for significant visual morbidity as a result. Achieving diagnostic competency is crucial because time is of the essence for higher risk categories of disease.

The second key finding is that plus disease was a prominent factor associated with misdiagnosis of category of disease, which is an important factor when determining type 2 and treatment-requiring disease. An internationally recognized reference image has been used to determine whether retinal findings noted on examination are consistent with the presence of plus disease.<sup>10</sup> This standard method of diagnosis lacks clear objectivity, and it has been demonstrated that there is a substantial degree of discrepancy even among ROP experts when diagnosing plus disease.<sup>11,12</sup> This is an area of concern given that the presence of plus disease warrants immediate treatment.<sup>11</sup> Indeed, our study confirms that ophthalmologists-in-training can routinely misdiagnose plus disease. The use of deep learning algorithms to facilitate plus disease diagnosis may be one way to improve diagnostic accuracy and guide management.<sup>10,13</sup> The Imaging and Informatics in Retinopathy of Prematurity (i-ROP) consortium has developed and validated a system built on deep convolutional neural networks that detects plus disease with 93% sensitivity and 94% specificity.<sup>10</sup> Furthermore, recent advancements in the i-ROP Deep Learning system have demonstrated the ability of artificial intelligence to objectively rate the severity of plus disease using a score,<sup>1-9</sup> potentially allowing for more precise disease surveillance.<sup>14,15</sup> The i-ROP Deep Learning system has been given breakthrough status by the U.S. Food and Drug Administration.<sup>16</sup>

The third key finding is that junior ophthalmologists-in-training are more likely to misdiagnose ROP compared to senior ophthalmologists-in-training. It is likely that part of the improvement in ROP diagnosis is from increasing experience rather

than proper didactic training. However, the rates of errors in diagnosing clinically significant ROP are still of concern. For example, although more than 60% of PGY-2 ophthalmologists-in-training misdiagnosed treatment-requiring ROP, 38% and 43% PGY-3 and PGY-4 ophthalmologists-in-training also misdiagnosed treatment-requiring ROP, respectively (**Figure 3**). Comprehensive ophthalmologists may carry the responsibility of screening for ROP due to the limited number of specialists available. Overall, it has been reported that 11% of ophthalmologists screen for ROP and not all are fellowship trained.<sup>17</sup> This shortage is in part due to geographic and population pressures, as well as concern over medical liability for ROP screening.<sup>5</sup> Because one-fifth of fellowship-trained ophthalmologists who were surveyed in one study intend to cease ROP management, it is paramount that residency programs graduate trainees competent in ROP examination.<sup>5</sup>

Incorporation of a tele-education system may supplement training in ROP by providing focused learning material, practice cases, and structured feedback.<sup>1</sup> This study is a subanalysis of a previous study that demonstrated the efficacy of a tele-education system for improving trainees' diagnostic competency.<sup>1</sup> This training system has also been piloted among ophthalmology trainees in Mexico who demonstrated significant improvement in their diagnostic specificity for type 2, treatment-requiring, and aggressive posterior ROP.<sup>1,18</sup> A tele-education program may be a valuable tool for enhancing the ability to identify clinically significant disease. This is also particularly beneficial in settings where screening is performed by a provider who is not an ROP expert, such as in a rural area, equipping the provider with the knowledge needed to identify cases of ROP that require urgent referral. Tele-education, as an instructional intervention, can be implemented into residency curricula in the first year to maximize trainees' knowledge of ROP screening in preparation for clinical rounds in the training years that follow. This will allow trainees to apply what they have learned from the web-based didactics to a practical clinical setting. Moreover, tele-triaging of patients and telemedicine may be particularly useful during circumstances such as the coronavirus disease 2019 (COVID-19) pandemic.<sup>19</sup> Thus, it is important that trainees obtain skills in screening for diseases, such as ROP, through various modalities, including digital image-based remote reading.

Web-based curricula can be used to promote diagnostic competency in telemedicine. Looking forward, the concept of automated diagnosis, such as through the i-ROP Deep Learning system, also supports the idea of automated methods to provide educational instruction tailored to the provider's level of expertise. Indeed, the trend toward artificial intelligence applications in a clinical setting may bridge the gap in providing specialized care in underserved regions. However, whether or not artificial intelligence systems are used for screening, if telemedicine for ROP care is implemented there are several important considerations. For example, training a workforce, setting up the infrastructure for device connectivity, and using a web-based platform are all key elements that need to be explored. The infrastructure required to establish an ROP program has been previously discussed by several investigators.<sup>20</sup> Still, further work is necessary to determine the feasibility of implementing an artificial intelligence-based program in regions with limited resources. Also, an analysis of the cost-effectiveness and program efficacy between traditional and artificial intelligence-based programs would provide valuable information.

There are several limitations to this study. First, performance of ophthalmologists-in-training was based on the analysis of responses from 32 trainees across six different sites. Two programs in particular were largely represented, and all of the programs were at institutions with both retina and pediatric ophthalmology fellowships. Trainees at the same program likely shared similar educational experiences, limiting the generalizability of our findings. The lack of standardization in ROP curricula suggests that trainees in other programs who did not participate may have varying degrees of exposure to ROP management. These trainees may service rural areas where they must meet the screening demands of the region, or they may train at institutions where ROP cases are managed by more specialized and experienced faculty. It must also be considered that although trainees at one program likely share similar experiences, there may still be variability in diagnostic accuracy. Although this variability may be a confounding factor, it may represent the variability seen in general clinical practice outside of a training setting. To more comprehensively assess the diagnostic performance of trainees, a wider selection of programs and a larger sample size will be needed for future work. In addition, administering a proctored examination would control access to outside educational material and likely increase the number of participants com-



pleting all of the cases. Overall, participation from six different programs in this study provides some insight into trainees' knowledge of ROP diagnosis that may guide future educational initiatives.

A second limitation is that the web-based system relies on wide-angle retinal images rather than indirect ophthalmoscopy.<sup>1</sup> The use of wide-angle fundus images standardizes the way the trainees view retinal findings, which enhances the educational experience but may hinder the clinical diagnostic process because trainees miss the opportunity to refine their skills in indirect ophthalmoscopy. However, color fundus photography is commonly used as part of the clinical evaluation, and there has been a shift in paradigm toward the use of tele-ophthalmology as a primary method for ROP screening and referral, particularly in low-resource settings. Thus, it is important to understand how trainees perform when diagnosing disease based on digital imaging.

The findings of this study contribute to our knowledge on the competency of ROP diagnosis by ophthalmologists-in-training in the United States and Canada. Given the rates of ROP misdiagnosis by ophthalmologists-in-training, the incorporation of ROP-focused education into residency curricula is critical to improve competency in ROP diagnosis and management. In addition, with the introduction of a revised international classification of ROP, web-based educational programs that can deliver updated curriculum will be beneficial for ophthalmology residents and fellows who may be responsible for ROP care in the future.<sup>21</sup>

## REFERENCES

- Chan RVP, Patel SN, Ryan MC, et al. The Global Education Network for Retinopathy of Prematurity (GEN-ROP): development, implementation, and evaluation of a novel tele-education system. *Trans Am Ophthalmol Soc.* 2015;113(T2):1-26. PMID:26538772
- Gilbert C. Retinopathy of prematurity: a global perspective of the epidemics, population of babies at risk and implications for control. *Early Hum Dev.* 2008;84(2):77-82. <https://doi.org/10.1016/j.earlhumdev.2007.11.009> PMID:18234457
- Cryotherapy for Retinopathy of Prematurity Cooperative Group. Multicenter trial of cryotherapy for retinopathy of prematurity: preliminary results. *Pediatrics.* 1988;81(5):697-706. <https://doi.org/10.1542/peds.81.5.697> PMID:2895910
- Early Treatment For Retinopathy Of Prematurity Cooperative Group. Revised indications for the treatment of retinopathy of prematurity: results of the early treatment for retinopathy of prematurity randomized trial. *Arch Ophthalmol.* 2003;121(12):1684-1694. <https://doi.org/10.1001/archophth.121.12.1684> PMID:14662586
- Wong RK, Ventura CV, Espiritu MJ, et al. Training fellows for retinopathy of prematurity care: a Web-based survey. *J AAPOS.* 2012;16(2):177-181. <https://doi.org/10.1016/j.jaaapos.2011.12.154> PMID:22525176
- Nagiel A, Espiritu MJ, Wong RK, et al. Retinopathy of prematurity residency training. *Ophthalmology.* 2012;119(12):2644-5.e1, 2. <https://doi.org/10.1016/j.ophtha.2012.07.015> PMID:23207022
- Myung JS, Paul Chan RV, Espiritu MJ, et al. Accuracy of retinopathy of prematurity image-based diagnosis by pediatric ophthalmology fellows: implications for training. *J AAPOS.* 2011;15(6):573-578. <https://doi.org/10.1016/j.jaaapos.2011.06.011> PMID:22153403
- Paul Chan RV, Williams SL, Yonekawa Y, Weissgold DJ, Lee TC, Chiang MF. Accuracy of retinopathy of prematurity diagnosis by retinal fellows. *Retina.* 2010;30(6):958-965. <https://doi.org/10.1097/IAE.0b013e3181c9696a> PMID:20168274
- Bakri SJ, Alniemi ST, Chan RV. Experiences of vitreoretinal surgery fellows in the United States. *Retina.* 2013;33(2):392-396. <https://doi.org/10.1097/IAE.0b013e31826b6700> PMID:23222495
- Brown JM, Campbell JP, Beers A, et al; Imaging and Informatics in Retinopathy of Prematurity (i-ROP) Research Consortium. Automated diagnosis of plus disease in retinopathy of prematurity using deep convolutional neural networks. *JAMA Ophthalmol.* 2018;136(7):803-810. <https://doi.org/10.1001/jamaophthalmol.2018.1934> PMID:29801159
- Chiang MF, Jiang L, Gelman R, Du YE, Flynn JT. Interexpert agreement of plus disease diagnosis in retinopathy of prematurity. *Arch Ophthalmol.* 2007;125(7):875-880. <https://doi.org/10.1001/archophth.125.7.875> PMID:17620564
- Hewing NJ, Kaufman DR, Chan RV, Chiang MF. Plus disease in retinopathy of prematurity: qualitative analysis of diagnostic process by experts. *JAMA Ophthalmol.* 2013;131(8):1026-1032. <https://doi.org/10.1001/jamaophthalmol.2013.135> PMID:23702696
- Ataer-Cansizoglu E, Bolon-Canedo V, Campbell JP, et al; i-ROP Research Consortium. Computer-based image analysis for plus disease diagnosis in retinopathy of prematurity: performance of the "i-ROP" system and image features associated with expert diagnosis. *Transl Vis Sci Technol.* 2015;4(6):5. <https://doi.org/10.1167/tvst.4.6.5> PMID:26644965
- Taylor S, Brown JM, Gupta K, et al; Imaging and Informatics in Retinopathy of Prematurity Consortium. Monitoring disease progression with a quantitative severity scale for retinopathy of prematurity using deep learning. *JAMA Ophthalmol.* 2019;137(9):1022-1028. <https://doi.org/10.1001/jamaophthalmol.2019.2433> PMID:31268518
- Gupta K, Campbell JP, Taylor S, et al; Imaging and Informatics in Retinopathy of Prematurity Consortium. A quantitative severity scale for retinopathy of prematurity using deep learning to monitor disease regression after treatment. *JAMA Ophthalmol.* 2019;137(9):1029-1036. <https://doi.org/10.1001/jamaophthalmol.2019.2442> PMID:31268499
- White F. Tech that detects cause of premie blindness gets federal nod. Published January 30, 2020. Accessed July 29, 2020. <https://news.ohsu.edu/2020/01/30/tech-that-detects-cause-of-preemie-blindness-gets-federal-nod>
- Kemper AR, Freedman SF, Wallace DK. Retinopathy of prematurity care: patterns of care and workforce analysis. *J AAPOS.* 2008;12:344-348. <https://doi.org/10.1016/j.jaaapos.2008.02.012> PMID:18440256
- Patel SN, Martinez-Castellanos MA, Berrones-Medina D, et al. GEN-ROP, i-ROP Research Consortium. Assessment of a tele-education system to enhance retinopathy of prematurity (ROP) training by international ophthalmologists-in-training in Mexico. *Ophthalmology.* 2017;124:953-961. <https://doi.org/10.1016/j.ophtha.2017.02.014> PMID:28385303
- Scanzer AC, Cole E, Valikodath N, et al. Implementation of COVID-19 protocols and tele-triage in an academic ophthalmology department. *J Acad Ophthalmol.* 2020;12(02):e151-e158. <https://doi.org/10.1055/s-0040-1715807>
- Al-Khaled T, Valikodath NG, Patel SN, et al. Addressing the third epidemic of retinopathy of prematurity through telemedicine and technology: a systematic review. *J Pediatr Ophthalmol Strabismus.* 2021;58(4):261-269. <https://doi.org/10.3928/01913913-20210223-01> PMID:34288773
- Chiang MF, Quinn GE, Fielder AR, Ostmo SR, Paul Chan RV, et al. International Classification of Retinopathy of Prematurity, Third Edition. *Ophthalmology.* 2021;8:S0161-6420(21)00416-4