

25 **SUMMARY** (200 words; max 200)

26 **Patient-reported outcome measures such as the Pediatric Eye Questionnaire (PedEyeQ) are**
27 **increasingly recognized as important in healthcare assessment. Defining normal PedEyeQ**
28 **thresholds would allow classification of individual children as having reduced versus normal**
29 **domain scores. We prospectively enrolled visually normal children (aged 0-17 years; n=310)**
30 **to calculate normal PedEyeQ domain thresholds. In addition, 48 children with bilateral visual**
31 **impairment (VI; best-eye acuity worse than 20/70, or 20/70 or better with limited visual fields)**
32 **were enrolled for validation. 5-17 year olds completed the Child PedEyeQ (4 domains).**
33 **Parents completed Proxy (5 domains) and Parent PedEyeQ (4 domains). Each domain was**
34 **Rasch scored (converted to 0-100) and normal thresholds defined as the 5th percentile of**
35 **scores in visually normal controls. Proportions of VI children with reduced scores were**
36 **calculated. For Child 5-11 PedEyeQ, 39% to 78% of VI children had reduced domain scores,**
37 **and 88% to 100% for 12- to 17-year-olds. For Proxy PedEyeQ, proportions ranged from 55%**
38 **to 100% and for Parent PedEyeQ \geq 83% had reduced scores. High prevalence of reduced**
39 **PedEyeQ domain scores in the VI cohort, validates the use of normal thresholds.**
40 **Nevertheless, variability in child self-reporting creates challenges for identifying individual 5-**
41 **11 year old children with reduced scores.**

42

43 INTRODUCTION

44 Previous studies have compared average Pediatric Eye Questionnaire (PedEyeQ)¹ domain
45 scores between patient cohorts (for example visual impairment,² specific eye conditions³) and
46 visually normal controls. Nevertheless, it would be helpful to define normal thresholds for PedEyeQ
47 domains, to allow classification of an individual child as having a reduced score. The aim of the
48 present study was to calculate normal thresholds for each PedEyeQ domain, and to validate the use
49 of thresholds in a clinical cohort with bilateral visual impairment (VI).

50 METHODS

51 Institutional Review Board approval was obtained from Institutional Review Boards at the
52 Mayo Clinic, Rochester, Minnesota and the University of Texas Southwestern Medical Center,
53 Dallas, Texas. All procedures and data collection were conducted in a manner compliant with the
54 Health Insurance Portability and Accountability Act and all research procedures adhered to the
55 tenets of the Declaration of Helsinki.

56 **Subjects**

57 *Visually Normal Controls*

58 Three-hundred-ten visually normal children were prospectively enrolled to calculate normal
59 thresholds: 104 aged 0-4 years, 104 aged 5-11 years and 102 aged 12-17 years (eTable 1). All had
60 no glasses or other refractive correction, no current or previous eye condition or treatment, and no
61 current diagnosis of learning disability, anxiety, depression, or a chronic health condition (allowing
62 only mild asthma, eczema, allergies and attention deficit disorder). Normal visual acuity in each eye
63 was defined as meeting age-referenced normal visual acuity thresholds,^{4,5} including Teller acuity
64 cards in very young children.⁶ For the majority, visual acuity was assessed in the office.
65 Nevertheless, due to the COVID-19 pandemic, we allowed home visual acuity testing in some
66 children aged 12 years and older (n=31 [30%]), using the Verana Vision smartphone application.⁷

67 248 (80%) of 310 visually normal control subjects have been reported in previous studies
68 comparing average values, but normal thresholds have not been previously reported.

69 ***Bilateral Visual Impairment***

70 Forty-eight children with bilateral VI, described in a previous publication evaluating
71 average PedEyeQ scores,² were included for the validation of normal thresholds: 22 aged 0-4 years,
72 18 aged 5-11 years and 8 aged 12-17 years. Visual impairment was defined using World Health
73 Organization criteria for moderate impairment, severe impairment or blindness: worse than 20/70 in
74 the better-seeing eye (in habitual refractive correction), or 20/70 or better with limited visual fields
75 (n=1) ([https://icd.who.int/browse11/l-](https://icd.who.int/browse11/l-m/en#/http%3a%2f%2fid.who.int%2fid%2fentity%2f1103667651)
76 [m/en#/http%3a%2f%2fid.who.int%2fid%2fentity%2f1103667651](https://icd.who.int/browse11/l-m/en#/http%3a%2f%2fid.who.int%2fid%2fentity%2f1103667651)).

77 **Pediatric Eye Questionnaire (PedEyeQ)**

78 Children >5 years completed the Child 5-11 or 12-17 year PedEyeQ comprising Functional
79 Vision, Bothered by Eyes/Vision, Social, and Frustration/Worry domains.¹ Parents completed the
80 Proxy PedEyeQ (0-4 years: Functional Vision, Bothered by Eyes/Vision, Social; 5-11 years and 12-
81 17 years: Functional Vision, Bothered by Eyes/Vision, Social, Frustration/Worry, Eye Care¹) and
82 Parent PedEyeQ (Impact on Parent and Family, Worry about Child's Eye Condition, Worry about
83 Child's Self-perception and Interactions, and Worry about Child's Functional Vision domains¹).

84 Questionnaires, scoring algorithms, and look-up tables are freely available at:

85 https://public.jaeb.org/pedig/view/Other_Forms.

86 **Analysis**

87 Rasch scores were calculated separately for each distinct PedEyeQ domain using look-up
88 tables (https://public.jaeb.org/pedig/view/Other_Forms) and converted to 0 (worst) to 100. The 5th
89 percentile of scores in normals was used to define thresholds for reduced domain scores.

90 Proportions with reduced scores in the VI cohort were calculated, with 95% confidence intervals.
91 SAS software version 9.4 (SAS Institute. Cary, NC) was used for all statistical analyses.

92 **RESULTS**

93 **Subjects**

94 Demographics of visually normal children are shown in eTable 1. For children with VI,
95 demographics and clinical characteristics have been previously reported.² Best-eye visual acuity
96 ranged from 20/107 (Teller acuity 5.6 cycles per degree) to light perception in 0-4 year olds, 20/80
97 to no light perception in 5-11 year olds and 20/20 (limited visual fields) to no light perception in 12-
98 17 year olds.

99 *Normal PedEyeQ thresholds*

100 Normal thresholds (5th percentile of scores) for Child, Proxy and Parent PedEyeQ domains
101 are shown in Table 1.

102 *Children with visual impairment*

103 Proportions with reduced scores in the VI cohort are shown in Table 2. Using the Child
104 PedEyeQ, 88% to 100% of scores were reduced for 12- to 17-year-olds and 39% to 78% for 5- to
105 11-year-olds (Table 2). Using the Proxy PedEyeQ, 55% to 100% of scores were reduced for 0- to 4-
106 year-olds, 61% to 94% for 5- to 11-year-olds, and 88% to 100% for 12- to 17-year-olds (Table 2).
107 Using the Parent PedEyeQ 83% to 100% of scores were reduced across age groups and domains
108 (Table 2).

109 **DISCUSSION**

110 In a large cohort of visually normal children, we calculated normal thresholds for each
111 PedEyeQ domain to enable identification of reduced domain scores in individual children. When
112 applying these thresholds in children with visual impairment we found the majority of children and
113 parents had reduced scores, providing validation of derived thresholds.

114 In most previous quality of life studies, comparisons between clinical and control cohorts
115 are reported as average values, allowing distributions of scores to be statistically compared with
116 each other. However a previous study in children applied normal thresholds,⁸ dichotomizing a
117 clinical cohort into those with normal versus reduced scores. Such an approach can be useful when
118 comparing domains or instruments with different items and/or scoring. It may also be useful to
119 apply normal thresholds longitudinally. Normal PedEyeQ domain score thresholds reported in the
120 present study may be useful for evaluating individual children in a clinical setting and can be
121 applied in clinical cohorts.

122 We found that normal Child PedEyeQ thresholds were lower in 5- to 11-year-old children
123 versus older children. Such lower scores might be expected because some young children have a
124 less realistic self-concept⁹ which may result in greater variability. When interpreting PedEyeQ
125 scores in 5- to 11-year-olds, it is important to be aware that lower normal thresholds make it more
126 difficult for a score to be categorized as reduced.

127 There are limitations to this study. The relatively low normal thresholds found in 5- to 11-
128 year-olds are driven by a small number of outlying scores and it is possible that these thresholds
129 may be higher if calculated using data from a larger cohort. Another limitation when applying a
130 normal threshold is that it may falsely dichotomize scores as “normal” versus “reduced,” due to
131 test-retest variability. Also, we did not evaluate proportions with reduced scores in moderate versus
132 severe visual impairment.

133 In summary, we have calculated normal PedEyeQ domain threshold scores and validated in
134 children with bilateral visual impairment. These thresholds can be applied in other clinical cohorts
135 to report proportions with reduced PedEyeQ scores. While we expect derived thresholds to perform
136 well in 12- to 17-year olds and for adults (Proxy and Parent raters), the relatively high variability of

- 137 domain scores in visually normal 5- to 11-year olds makes it more challenging to identify
- 138 individuals with reduced scores in this age group.

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172 **eTable 1.** Demographics of Visually Normal Controls.

	Visually normal subjects (n=310)		
	0-4 year olds (n=104) N (%)	5-11 year olds (n=104) N (%)	12-17 year olds (n=102) N (%)
Sex of child: female	56 (54%)	52 (50%)	51 (50%)
Median age (years)	2	7	13
Race/ethnicity			
White	62 (60)	71 (68)	69 (68)
Hispanic/Latino	10(10)	5 (5)	2 (2)
American Indian / Alaskan Native	1 (1)	0 (0)	0 (0)
Asian	12 (12)	11 (11)	19 (19)
Black/African American	2 (2)	5 (5)	4 (4)
More than one	15 (14)	9 (9)	6 (6)
Other	2 (2)	3 (3)	2 (2)

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11- PedEyeQ Normal Thresholds

175 **Table 1.** PedEyeQ Thresholds Derived From Visually Normal Children and Their Parents to Define Reduced Domain Scores (Below
 176 the 5th Percentile).
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Thresholds for reduced domain scores						
Child	N	Functional Vision	Bothered by Eyes/Vision	Social	Frustration / Worry	
5-11	104	70.0	75.0	65.0	60.0	
12-17	102	90.0	95.0	100	83.3	
Proxy	N	Functional Vision	Bothered by Eyes/Vision	Social	Frustration / Worry	Eye Care
0-4	104	100	100	94.4		
5-11	104	90.0	95.0	100	100	100
12-17	102	95.0	100	100	100	100
Parent	N	Impact on Parent and Family	Worry about Child's Eye Condition	Worry about Child's Self Perception and Interactions	Worry about Child's Functional Vision	
0-4	104	100	90.0	100	100	
5-11	104	100	90.0	100	93.7	
12-17	102	100	90.0	100	100	
Overall	310	100	90.0	100	93.8	

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12- PedEyeQ Normal Thresholds

179 Table 2: Proportions of Visually Impaired Subjects Meeting Criteria for Reduced Scores (below the 5th percentile in normal controls),
 180 on each PedEyeQ domain.

Proportions with Reduced Domain Scores						
Child	N	Functional Vision	Bothered by Eyes/Vision	Social	Frustration / Worry	
5-11	18	78%	67%	44%	39%	
12-17	8	100%	88%	88%	100%	
Proxy	N	Functional Vision	Bothered by Eyes/Vision	Social	Frustration / Worry	Eye Care
0-4	22	100%	100%	55%	NA	NA
5-11	18	94%	83%	72%	61%	61%
12-17	8	100%	100%	100%	88%	100%
Parent	N	Impact on Parent and Family	Worry about Child's Eye Condition	Worry about Child's Self Perception and Interactions	Worry about Child's Functional Vision	
0-4	22	95%	100%	95%	100%	
5-11	18	89%	100%	83%	94%	
12-17	8	100%	100%	100%	100%	
Overall	48	94%	100%	92%	96%	

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