CVD-AES: Construct and concurrent validity of a novel scale to measure the adverse effects of congenital colour vision deficiency

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In recent decades, colour has become an increasingly relied-upon tool within the classroom, but, little research has been conducted on the impact of this change on the educational experiences and outcomes of colour-vision deficient (CVD) students. Here, following a qualitative study of childhood impacts of CVD, we developed and validated a questionnaire (the CVD Adverse Effects Scale; CVD-AES) measuring the adverse effects of CVD, focusing on education. CVD participants reported significantly more difficulties than control participants in total and across all subscales: Education (15 items), Social (5 items), Emotions (13 items) and Day-to-Day (9 items), demonstrating construct validity and providing evidence that CVD has an adverse effect on affected individuals. There was a significant correlation between scores on the CVD-AES and a comparable measure, the Colour Blind Quality of Life (CBQoL) scale, demonstrating good concurrent validity of the novel scale. We suggest that the questionnaire should be used alongside robust diagnostic tools for CVD, allowing investigation of the effect of CVD type and severity, and to understand the impacts of the increasing reliance of colour on CVD students across a range of educational contexts.

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Introduction

For most people, colour vision arises from the responses of three types of colour-sensitive cells (cone photoreceptors) in the eye: one sensitive to short-wavelength light (S-cones), one sensitive to middle-wavelength light (M-cones) and one sensitive to long-wavelength light (L-cones). X-linked colour-vision deficiency (CVD) is a hereditary condition affecting 8% of males and 0.4% of females involving one of the latter two cone types. In anomalous trichromacy, the milder form, either the M-cones (deuteranomaly) or the L-cones (protanomaly) are spectrally shifted in the wavelengths of light they optimally respond to. In dichromacy, the more severe form, either the M-cone (deuteranopia) or L-cone (protanopia) is missing. Depending on type and severity, CVD individuals' ability to discriminate between colours can be significantly compromised. The impacts of CVD often include issues at work, problems watching and participating in sports, difficulties interpreting colour-coded information, and learning at school [1].

Barry and colleagues have developed a questionnaire (the CBQoL) to measure quality of life in those with CVD, including sections on health and lifestyle, emotions, and work [2]. The questionnaire was developed following literature searches, a focus group with six CVD individuals and two individuals with normal colour vision (NCV), and an expert focus group with three consultant ophthalmologists and the author, who is himself CVD. Following development, the questionnaire was distributed to 65 CVD and 332 NCV participants and validated against other measures of quality of life: the Short Form 36 [3] measuring health-related quality of life, and the Positive State of Mind Index [4]. CVD individuals reported significantly worse quality of life overall and in each section than controls with normal colour vision. However, the CBQoL does not measure the pertinent impact of CVD on education, and as such is primarily focused on adults, furthered by its inclusion of the impacts on career.

A significant impact of CVD on school achievement has been found in general [5] and specific subjects including languages, mathematics, science and geography [6]. CVD children have also reported significantly more difficulties with colour-related tasks at school than their peers with normal colour vision [7-8].

Despite the high prevalence of the condition and clear evidence of its adverse effects on education, CVD is not routinely screened for, meaning those affected, particularly anomalous trichromats, may remain undiagnosed, unable to develop educational coping strategies and plan suitable career options. Formal screening currently only occurs in seven states in the USA [9], and there is no formal screening in India [10], or, as of 2009, the UK (Department of Health, 2009 [11]). Furthermore, in the UK CVD is not classed as a special educational need (SEN), meaning there is no legislation requiring reasonable adjustments in educational settings when a diagnosis is obtained. In England, a school-aged child or young person is classed as having a SEN if he or she "has a disability that prevents or hinders him or her from making use of facilities of a kind generally provided for others of the same age in mainstream schools or mainstream post-16 institutions" (the Children and Family Act, UK, Section 20 (2b), 2014 [12]); and a person is defined as having a disability if they have "(a) a physical or mental impairment, and (b) the impairment has a substantial and long-term adverse effect" on their "ability to do normal daily activities" (the Equality Act, UK, Section 6 (1), 2010 [13]). It is reasonable to assume that a number of children with CVD will have difficulties making use of colour-based facilities and learning materials in schools which are accessible to their peers with normal colour vision. What is less clear is whether these difficulties will have a "substantial and long-term adverse effect". Nonetheless, there is ambiguity surrounding CVD's status as a disability (Women and Equality Committee, 2020 [14]). UK case law has not recognised CVD as a disability for the purposes of the Equality Act (2010) but this relates to a specific adult male's ability to cook, read/interpret documents/text, and watch sport (Bessell v. the Chief Constable of Dorset Police [15]). As such, CVD as a disability in the UK education system remains unchallenged. Establishing whether CVD in the classroom has a 'more than minor or trivial' adverse and long-term impact on education is key to move beyond the current status quo.

To summarise, there is a need to provide further evidence of the impacts of CVD on education and to consider how the outcomes relate to important real-world challenges in the lives of CVD children in school. The present study builds on our previous, qualitative data [9-10] and reports the development of a new scale (CVD-AES) that will allow researchers to gather quantitative data on the impacts of CVD in a wider range of affected individuals.

Study 1: construct validity

Methods

Participants

Participants were recruited through an online link posted on social media websites e.g. Facebook and Twitter. Six CVD participants were removed for self-reporting total or blue-yellow colour-blindness. One control participant was removed for entering the same answer ("4: daily") for every question, including distractor items. Participants were asked to report their colour-vision status (NCV or CVD) and their CVD subtype, if known. Of the remaining 425 participants there were 154 NCV controls (mean age=35.41; SD=19.67; 92 females) and 271 CVD participants (mean age=33.55; SD=19.46; 16 females). There were 82 anomalous trichromats (55 deuteranomalous and 27 protanomalous); 79 dichromats (41 deuteranopes and 38 protanopes) and 110 participants could not specify their subtype. 337 participants were from the UK, with the remaining participants from 11 other countries in Europe, five in Asia, two in North America, two in South America, two in Oceana, and one in Africa.

Scale development

The CVD-Adverse Effects Scale (CVD-AES) was developed using the results of a thematic analysis of interviews and focus groups with 26 CVD adults, 11 CVD children and 12 parents of CVD children, focusing on the experience of growing up with CVD [16-17]. Specifically, it was developed from the themes/subthemes: barriers to learning within education, negative reactions of others, negative impacts on emotions and self-view, and a day-to-day subtheme.

Subscale	Prompt	Items
Education	Think about your experiences at school in the past. How often have you had problems in the lessons or tasks below because of your colour vision?	English (including literature and language), Maths, Science (including biology, chemistry, physics and electronics), Geography, History, Foreign languages (e.g. French, Spanish, German), Art, PE, Food technology, IT, Music, Reading information from the board, Setting learning targets, objectives or aims to improve on, Telling the teacher what you have found easy or hard, Answering questions in tests or exams
Social	Think about your experiences with other people in the past. How often have each of these things happened to you?	People thinking you are stupid because of problems with your colour vision, People thinking you are lazy because of problems with your colour vision, People not believing what you tell them about your colour vision, People (who are not healthcare professionals) testing your colour vision, for example asking you to name colours or tell the difference between coloured objects like pencils, People laughing at you for making mistakes about colour
Emotions	How often have you felt the following ways because of your colour vision?	Frustrated, Special/Unique, Embarrassed, Excited, Worried, Sad, Like a nuisance, Proud, Alone, Self-conscious, Angry, Happy, Like the odd-one-out
Day-to-day	Think about your everyday experiences. How often have you had problems with the tasks below because of your colour vision?	Using technology such as charging devices, WiFi routers and TV boxes, Doing physical hobbies such as sports, Doing arts and crafts, Using software on computers and devices such as playing games, Cooking food, Choosing fruit and vegetables, Eating food that looks unappealing, Understanding information on labels, graphs and maps, Finding clothes that match

Table 1: Subscales, prompts and items for the CVD-AES.

The CVD-AES comprises of 42 items (Table 1): 15 on Education, 5 on Social impacts, 13 on Emotions (e.g. "Frustrated"; including 4 distractor positive emotions), and 9 on Day-to-day impacts. Participants were asked to indicate how often they experienced specific difficulties on a 5-point Likert scale ranging from "0: Never" to "4: Daily". For the Education and Day-to-day items there was also a "Not relevant to me" response option, for example, if they had not studied the subject or encountered the day-to-day task in question.

Data analyses

The four distractor, positive emotion, items ("Special/unique", "Happy", "Excited" and "Proud) were removed, so that impact score reflected the negative impact of CVD only.

As participants were able to answer "Not relevant to me" for some questions, the impact score needed to reflect how many questions were answered. Thus, the actual impact score was expressed as a proportion of total possible impact (ranging from 0 to 1). The highest impact score per item was 4, therefore impact score was calculated using the following formula:

$$Impact\ score = \frac{\Sigma\ Given\ answsers}{Number\ of\ questions\ answered \times 4} \quad (1)$$

When examining Education and Day-to-day subscales, any participant who answered "Not relevant to me" to over 50% of subscale questions was removed.

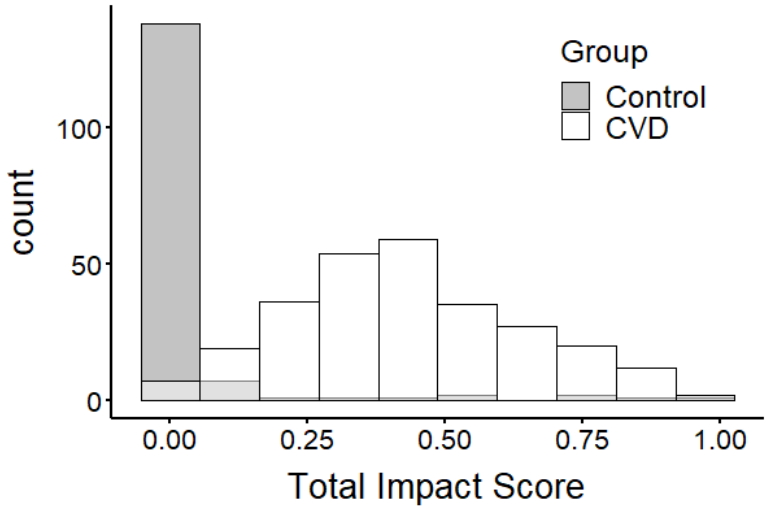


Figure 1: Distribution of total impact scores for control and CVD participants.

Shapiro-Wilk tests revealed that neither the impact scores for controls nor CVDs were normally distributed for Total impact score (see Figure 1) or any subscales (see Table 2). Construct validity of the CVD-AES, i.e. whether the scale truly measures the adverse effects of CVD, was assessed through known group validity; determining whether the scale can discriminate between groups (i.e. controls and CVDs) known to differ on the variable of interest (adverse effects of CVD). Wilcoxon Signed Ranks tests were carried out to investigate whether CVD participants scored significantly higher on the questionnaire than controls.

	Control		CVD	
	W	P	W	P
Total	0.310	<0.001	0.985	0.009
Education	0.360	<0.001	0.974	<0.001
Social	0.293	<0.001	0.967	<0.001
Emotion	0.247	<0.001	0.934	<0.001
Day-to-day	0.261	<0.001	0.982	0.002

Table 2: Results of Shapiro-Wilk tests for normality for control and CVD groups on total impact score and subscale impact scores.

All data analyses were conducted in R-Studio.

Results

A Wilcoxon Signed Rank test revealed that CVD participants had significantly higher Total impact scores than control participants (W=39805, p<0.001; see Table 3 for medians). This significant difference is illustrated in Figure 2, showing the median scores of control participants at zero (grey bars). Figure 2 also displays potential outliers in the control group (grey dots). The Inter Quartile Range (IQR) criterion identified 38 potential outliers within the control group, i.e. those with an impact score larger than the 75th percentile + 1.5(IQR). See Discussion for consideration of why these outliers may have occurred.

Total/Subscale	Statistic	Control	CVD
Total	Median	0.000	0.413
	IQR	0.000	0.283
Education	Median	0.000	0.393
	IQR	0.000	0.348
Social	Median	0.000	0.400
	IQR	0.000	0.350
Emotions	Median	0.000	0.306
	IQR	0.000	0.389
Day-to-day	Median	0.000	0.528
	IQR	0.000	0.367

Table 3: Medians and interquartile ranges (IQR) for Total and subscale impact scores for control and CVD participants.

Further Wilcoxon Signed-Rank tests determined whether a group difference was present across all subscales. Impact scores for the subscales were calculated in the same way as total impact score (see Data Analyses). CVD participants scored significantly higher on all subscales than controls (Tables 3 and 4). Figure 2 illustrates these differences as well as showing potential outliers in the control group for each subscale.

			Control		CVD	
	W	***p***	**n**	**Median**	**n**	**Median**
Education	25568	<0.001	110	0.000	251	0.393
Social	39333	<0.001	154	0.000	271	0.400
Emotion	39199	<0.001	154	0.000	250	0.306
Day-to-day	33630	<0.001	132	0.000	268	0.528

Table 4: Results of Wilcoxon Signed Rank Tests comparing subscale impact scores for control and CVD participants.

Within the control group, the IQR criterion identified the following potential outliers in each of the subscales: 21 in Education, 21 in Social, 17 in Emotions and 11 in Day-to-day.

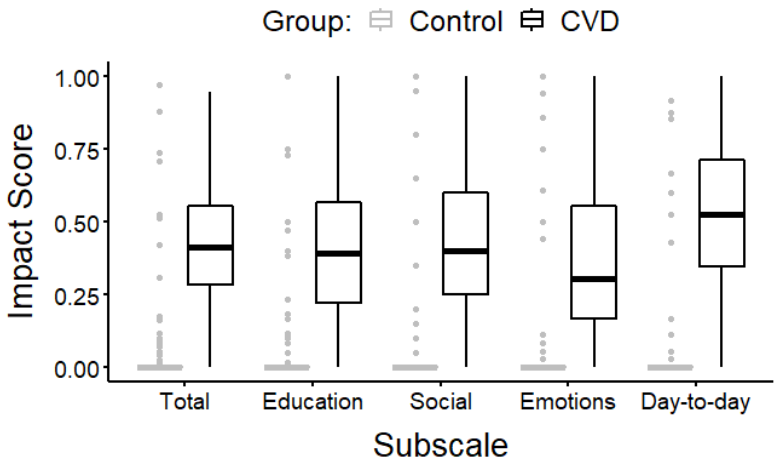


Figure 2: Comparison of Total and subscale impact scores of controls (grey) and CVD (black) participants. Medians are indicated by bold horizontal bars.

Study 2: concurrent validity

Methods

Participants

As before, participants were recruited through an online link posted on social media websites. Only self-reported CVD individuals were recruited, and they were asked to report their CVD subtype, if known. One participant was excluded due to total colour-blindness. Of the remaining 74 participants (mean age=44.35; SD=18.53; 4 females) there were 23 anomalous trichromats (17 deuteranomalous, 6 protanomalous); 20 dichromats (10 deuteranopes, 10 protanopes) and 31 participants who could not specify their subtype. 64 participants were from the UK, four from North America, two from Europe and one from South America.

Materials

Characteristics of the CVD-AES are described in the previous section.

In order to test concurrent validity, responses were compared to responses to the Colour Blind Quality of Life (CBQoL) scale [2]. The CBQoL is a 23-item questionnaire which assesses the impact of CVD on quality of life (QoL) by asking about the severity of problems respondents face within three subscales; Health and Lifestyle (e.g. "Difficulty choosing or buying clothes"), Emotions (e.g. "Feeling anxious because of issues caused by problems seeing colours") and Work (e.g. "Being limited in choice of work or career"). A 6-point Likert scale ranging from "1: A severe problem" to "6: No problem" is used, thus scores range from 1 to 6, and a lower score indicates worse QoL. Participants are also given the option to respond "not applicable" to each item.

Data analyses

The CVD-AES was scored as per the previous data analyses section.

The CBQoL was scored according to personal communication with the authors; means were generated for each of the subscales (Health and Lifestyle, Emotions, and Work). Where data were missing (i.e. the participant has responded "not applicable"), the missing data point was replaced with the mean of their remaining responses, as long as at least 66% of the remaining items had a valid numeric response. In order to compare Total Impact from the CVD-AES with the CBQoL, a total mean CBQoL score was also calculated.

Shapiro-Wilk tests revealed that the impact scores for the CVD-AES (W=0.977, p=0.209) and the CBQoL (W=0.972, p=0.095) were normally distributed. A Pearson's correlation was carried out to determine concurrent validity (the amount of agreement between a novel and an established scale measuring similar constructs) by investigating the strength of the relationship between the two measures.

To investigate whether the CVD-AES is better suited to younger participants, the number of "NA" responses to items on the CVD-AES and the CBQoL were compared between adults (18 years and over) and children (under 18 years). Number of "NA" responses to the CVD-AES were not normally distributed for children (W=0.727, p=0.003), or adults (W=0.873, p<0.001). Number of "NA" response to the CBQoL were normally distributed for children (W=0.940, p=0.581), but not adults (W=0.612, p<0.001). Therefore, Wilcoxon Signed Rank tests were carried out to investigate these potential differences.

Results

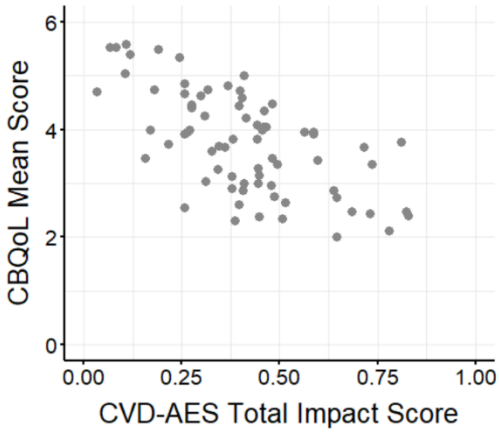
To determine concurrent validity, the relationship between responses to the CVD-AES and the CBQoL was investigated. Descriptive data for the novel scale and the CBQoL and their subscales are shown in Table 5.

Scale	Total/Subscale	Mean	SD	Minimum	Maximum
CBQoL	Total	3.746	0.945	2.000	5.591
	Health and Lifestyle	3.390	1.051	1.000	5.818
	Emotions	4.207	1.184	1.667	6.000
	Work	3.667	1.324	1.000	6.000
CVD-AES	Total	0.410	0.186	0.033	0.829
	Education	0.376	0.206	0.000	0.786
	Social	0.436	0.229	0.050	1.000
	Emotions	0.341	0.231	0.000	0.917
	Day-to-day	0.518	0.238	0.000	1.000

Table 5: Mean, standard deviation (SD), minimum and maximum scores on the CBQoL and CVD-AES, and their subscales.

A Pearson's correlation found a moderate, but significant negative correlation between the two measures; $r(72)$=-0.644, p<0.001. The relationship between the scales is presented in Figure 3.

Children gave significantly more "NA" responses to the CBQoL (median=5) than adults (median=0, W=27.00, p<0.001), however no difference was found for "NA" responses to CVD-AES items (W=370.50, p=0.189; Adults median=2, Children median=1). Furthermore, a paired Wilcoxon Signed Rank test found children submitted significantly more "NA" responses to the CBQoL than to the CVD-AES (V=36.00, p=0.014). These results are visualised in Figure 4.



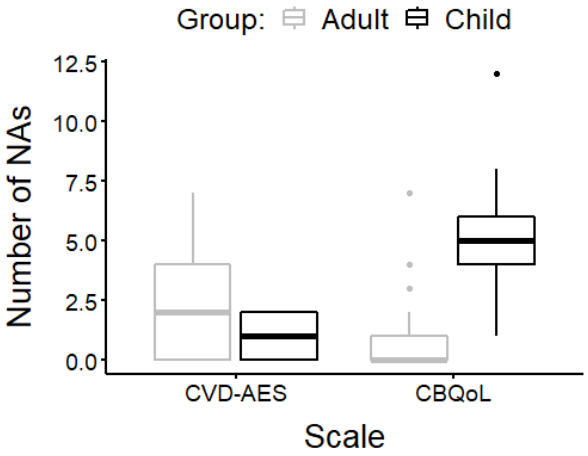


Figure 3 (left): CBQoL mean scores plotted against CVD-AES total scores.

Figure 4 (right): Number of "NA" responses submitted by adults (grey) and children (black) to the CVD-AES and the CBQoL.

Discussion

The present studies demonstrate the construct/known-group and concurrent validity of the CVD Adverse Effect Scale (CVD-AES), a novel scale to measure the impact of CVD, with a focus on education. CVD participants scored significantly higher than controls on the overall questionnaire and on each subscale, demonstrating that the CVD-AES does indeed capture the impacts of the condition, and differentiates between CVD and NCV individuals, validating the impact of CVD on education, social situations, emotions and day-to-day tasks. In addition, a moderate but significant negative correlation between mean scores on the CVD-AES and mean score on the comparable CBQoL were found. As low scores on the CBQoL indicate a lower QoL due to CVD, and higher scores on the CVD-AES indicate higher negative impact of CVD, this correlation is in the expected direction.

The correlation demonstrates that the novel scale is a valid measure of the impact of CVD, as it corresponds to a related measure of the impact of CVD on QoL. However, as the correlation was only moderate, it also demonstrates that the two measures are somewhat different. This is entirely expected as the CVD-AES's focus is on education, rather than the adult impacts of CVD. Child participants submitted significantly more "not applicable" responses to the CBQoL than adults, however the same was not found for the CVD-AES, suggesting that items on the novel scale are more applicable to children, giving credit to the CVD-AES as a valid, independent measure.

One limitation of this study was the use of self-report of CVD. While this was necessary to allow the scale to be disseminated online, it means the grouping of participants into CVD and control groups is uncertain. Furthermore, it was not possible to ascertain whether participants who could not report their CVD subtype were not suffering from an acquired CVD, rather than congenital CVD. Use of the scale alongside detailed categorisation of participants, for example using Rayleigh matches, would provide more certainty, and would allow a more in-depth analysis, such as investigating the impacts of different types and severities of CVD. Related to this limitation are the presence of outliers in the control group. It is possible these outliers may have undiagnosed CVD. Albany-Ward and Sobande report that 80% of UK CVD schoolchildren arrive in Year 7 without a diagnosis, meaning that, unless a diagnosis is sought out after this point, their CVD will remain undiagnosed [18]. Furthermore, it is possible that other underlying vision problems were present within these outliers as there was no question in the demographics section asking about other vision problems. This should be included in the future.

The findings regarding education support previous studies reporting a significant negative impact of CVD on education [5,7-8], as we find CVD participants report more frequent issues at school due to their colour vision. This also questions CVD's lack of status as an SEN in the UK, as we demonstrate here that CVD individuals report significantly higher impacts on every CVD-AES subscale. This demonstrates the adverse effects of CVD and suggests that affected students are unable to make use of the facilities and resources which are accessible to their NCV counterparts.

The CVD-AES can not only be used to determine the impact of an individual's CVD (e.g. for use by a teacher to provide reasonable adjustment to CVD students), but also to investigate other factors involved in CVD impact. For example, it is probable that the educational impacts of CVD are higher for today's children than they were in the past, due to the increase in reliance on coloured teaching materials. For instance, it is now common practice in UK schools to mark books with coloured bands to denote reading age or level, ask children to indicate task difficulty using traffic light colours and use coloured educational tools like Unifix or Numicon [19]. Despite the recent increase in the use of colour, the termination of routine testing in UK schools in 2009 was likely linked to the publication of a cohort study of individuals born in 1958, attending school in the 1960s-70s [20]. The study reported no impact of CVD on educational outcomes and concluded that CVD confers no functional disadvantage in an educational context. The present questionnaire, alongside data reported elsewhere [5-6], can be used to strongly challenge their conclusion in today's educational environment.

Conclusions

In conclusion, the present studies demonstrate good construct and concurrent validity for a novel measure of the potential impacts of CVD, the CVD Adverse Effects Scale (CVD-AES). CVD individuals report significant adverse effects of the condition overall, and on all subscales. Responses to the novel scale significantly correlate with responses to an established, validated measure of the impact of CVD on quality of life (the CBQoL). The CVD-AES can be used in the future to provide further evidence that CVD can present as a significant barrier to learning, and therefore, should be screened for in schools, and when present, appropriately accommodated for.

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