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published in
Infant Behavior and Development
2021

DOI (link to publisher)
10.1016/j.infbeh.2021.101585

Infant Behavior and Development 64 (2021) 101585

Contents lists available at ScienceDirect

Infant Behavior and Development

journal homepage: www.elsevier.com/locate/inbede

Identifying when children with visual impairment share attention: A novel protocol and the impact of visual acuity

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ARTICLE INFO

Keywords:
Joint engagement
Joint attention
Shared attention
Visual impairment
Low vision
Blindness

ABSTRACT

Background: In Coordinated Joint Engagement (CJE), children acknowledge that they and their social partners are paying attention to the same object. The achievement of CJE, critical for healthy development, is at risk in infants with visual impairment (VI). Research on CJE in these children is limited because investigators use a child's gaze switch between social partner and object to index CJE. Research is needed that identifies CJE in children with VI using behaviors that do not require normal vision and that explores the relationship between CJE and visual function. This study aimed to (a) develop a protocol for identifying CJE in children with VI, and (b) explore the relationship between CJE and infants' visual acuity (VA) and contrast sensitivity (CS), measured with Preferential Looking (PL) techniques and Visual Evoked Potential (VEP).
Methods: A protocol that included 9 indices of CJE that did not require normal vision was developed to code videos of 20 infants with VI (mean age =1 year, 6 months, 27 days) and their caregivers. The percentage of CJE episodes in which each index was observed was calculated. Inter-coder reliability was measured using Cohen's Kappa. Linear regression analysis was used to examine the relationship between the infants' visual function and CJE.
Results: Inter-rater reliability between a first coder and each of two second coders were 0.98 and 0.90 for determining whether the child participated in CJE. The following indices were observed the most (in 43–62 % of CJE): child's body orientation to caregiver, gaze switch between caregiver and object, and vocalization to caregiver. The only significant model included VA (measured with PL) as a single predictor and explained 26.8 % of the variance in CJE.
Conclusions: The novel protocol can be used to identify CJE in children with VI with good inter-coder reliability. The data suggest that children with lower VA exhibited less CJE.

Abbreviations: CJE, coordinated joint engagement; VI, visual impairment; VA, visual acuity; CS, contrast sensitivity; PL, preferential looking; VEP, visual evoked potential; ASD, Autism Spectrum Disorder; CJE%, percentage of joint engagement coded as coordinated; CJE% Toys 1, CJE measured using infants' toys; CJE% Toys 2, CJE measured using standard set of toys.

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https://doi.org/10.1016/j.infbeh.2021.101585
Received 11 September 2020; Received in revised form 18 May 2021; Accepted 18 May 2021
Available online 29 May 2021

1. Introduction

Early in development, infants pay attention to either a social partner or an object, but not to both, within the same interaction (Adamson & Mc Arthur, 1995). With the emergence of "joint engagement", around 12 months of age, infants start to coordinate their attention between a social partner and an object of mutual interest, effectively "sharing" their focus of attention with the social partner (Bakeman & Adamson, 1984). In "coordinated" joint engagement (CJE), the infant acknowledges the participation of the social partner in the shared attention, for instance by looking back and forth between the social partner's face and the object (this is called "joint attention" by authors like Tomasello & Carpenter, 2007). In "supported" joint engagement, the infant does not show this acknowledgement and the shared attention relies more on the social partner's scaffolding (Adamson, Bakeman, Deckner, & Nelson, 2012).

CJE is associated with positive developmental outcomes in language (Adamson, Bakeman, & Deckner, 2004), conventional use of objects (Bigelow, Maclean, & Proctor, 2004), theory of mind (Charman et al., 2000), and emotional regulation (Van Hecke et al., 2012). Furthermore, "atypical" CJE (i.e., outside the developmental norm in sighted children) is an early sign of Autism Spectrum Disorder (ASD; Hobson, 2005). Childhood visual impairment (VI) introduces challenges to CJE development (Dale & Salt, 2007). For instance, compared to sighted peers, children with VI show a slower development of reaching after hearing an object's sounds (Warren, 1984), and of locomoting independently, which is conducive to encountering objects (Brambring, 2006). Consequently, children with VI may engage less with objects, which leads to fewer opportunities for caregivers to interpret these children's behaviors as related to an external focus (Bigelow, 2003). Furthermore, caregivers can have difficulties identifying when these children attend to an object because of the unusual ways they may do so, e.g., by turning their head away from the object while listening (Preisler, 1991). Despite the importance of CJE and its development being at risk in children with VI, there is not enough research to determine the normative development of CJE in these children. This knowledge is needed to inform intervention practices that promote healthy development, and to determine when CJE is *truly* atypical (i.e., compared to the norms of children with VI) and a sign of ASD (Pérez-Pereira & Conti-Ramsden, 2005).

The limited research on CJE in children with VI is in part due to this population's low incidence, with those without additional disabilities being extremely rare. A US study that included 29 states reported 2,155 children with VI. Of these only 694 had no additional disabilities (Hatton, Schwietz, Boyer, & Rychwalski, 2007). Additionally, this research has been discouraged by the fact that investigators primarily index CJE using an infant's gaze switch between social partner and object, although some authors have acknowledged the limitation of this approach (Adamson et al., 2004; Carpenter, Nagell, Tomasello, Butterworth, & Moore, 1998; Yu & Smith, 2013). Consequently, CJE research in children with VI includes very small samples (Bigelow, 2003) or indirect measures (e.g., parental reports) rather than direct observation of children's behaviors (Dale, Tadić, & Sonksen, 2013). However, CJE can be established without involving the infant's gaze—e.g., children can feel their hands and the parents' hands exploring the same object (Bigelow, 2003). And although the gaze switch behavior is not possible for children who are blind, it is possible for children with VI that have sufficient residual vision, particularly if the index does not require eye-to-eye contact with the social partner. Therefore, it is necessary and possible to find ways to index CJE in children with VI using behaviors that do not require normal vision.

Studies have shown that children with more severe VI (e.g., profound versus severe) are associated with lower scores on CJE measures (Dale et al., 2013; Preisler, 1991). Less known is the impact impairment of different visual functions, specifically visual acuity (VA) and contrast sensitivity (CS), has on CJE. VA is the ability to see fine detail and is measured with tasks such as reading letters of decreasing size or seeing stripes of decreasing width. CS is the ability to detect differences in brightness (i.e., subtle shades of gray) and is relevant for tasks like seeing stairs and curbs, and detecting the faint shadows in faces that carry the visual information related to facial expressions (Orel-Bixler, 2014). There is evidence that for predicting infants with VI's participation in joint engagement, *without* differentiating between coordinated and supported, CS is a better predictor than VA (Urqueta Alfaro, Morash, Lei, & Orel-Bixler, 2018). However, it is unknown which of these visual functions would best predict these infants' participation exclusively in CJE, that is, only considering joint engagement episodes in which the child acknowledges the role of the caregiver in the shared attention.

This study aimed to: (a) develop and test a coding protocol for identifying CJE in infants with VI, (b) investigate whether CS or VA is a better predictor of CJE. Given the relevance of CS for seeing objects and facial expressions we predicted that CS would be the better predictor.

2. Methods

This study was approved by the Committee on Research Ethics of the Université de Montréal (18-116-CERES-D). The data were collected for a previous study and the detailed methodology has been reported elsewhere (Urqueta Alfaro et al., 2018). In what follows, we present a summary of the most relevant methodological aspects.

2.1. Participants

Twenty infants with VI (*M* age =1 year, 6 months, 27 days, *SD* age =3 months, 6 days; 10 girls) without other disabilities, and their main caregivers (*M* age = 31 years, *SD* age = 7 years; 18 biological mothers, 1 adoptive mother, 1 father) participated in this study. Infants' VI was confirmed by an optometrist (see 2.2.2. visual measures). No infant was assessed as having no light perception (blind). Thus, the sample includes only VI infants with residual vision and does not include blind infants. Table 1 shows infants' ages and visual diagnoses.

Six infants were White, 5 Hispanic, 2 Asian, 1 African American, and the remaining 6 were of more than one race/ethnicity. The caregivers' educational levels varied; 3 had a master's degree, 5 had a bachelor's degree, 8 had more than a high-school diploma (e.g., technical school), 2 had a high-school diploma, and 2 had completed middle school. Based on information provided by the caregivers, four families in the sample had a per capita income (i.e., income divided by the number of people living at the family's home) below $5,000; 4 between $8,750 and $15,833; 5 between $18,750 and $20,833; 2 had $23,750; and 2 had $31,667. All caregivers received intervention services to support their children's development and provided informed consent prior to study procedures.

2.2. Measures

2.2.1. Coordinated joint engagement

Caregiver-infant interactions were video recorded during naturalistic play for 30 min at their homes. Infant and caregiver could be at any distance from one another, as long as they remained within the viewing range of the video cameras. The toys were placed between child and caregiver. Given that children with VI perform best with familiar toys, during the first 15 min dyads used the children's own toys. To counteract differences due to socioeconomic level, a standard set of toys was used during the last 15 min. This study utilized the portions of the video data that were coded as episodes of joint engagement in a previous study. Joint engagement episodes met all three of the following conditions: (a) The infant was engaged with the same object/event with which the caregiver was engaged. Engagement was indexed when an infant was attending with any sense(s), i.e., gazing, hand manipulation, body orientation or performing an action with an object. (b) The caregiver acknowledged the infant's participation in the activity (i.e., attending to the infant's engagement with the object). For example, child and caregiver rolled a ball back and forth between them. But, if the child rolled the ball toward the caregiver, the ball touched the caregiver and as a result bounced back to the child while the caregiver was not attending to the child, join engagement was not coded. (c) The caregiver's involvement with the object influenced the child's experience with such object by calling the infant's attention to it, and/or acting on it in a way that influenced the infant's experience of the object. For example, the caregiver shows her infant a pompom, whereupon the child attends to it. Or, the caregiver starts shaking a pompom the child was mouthing, thus changing the infant's experience of the pompom from attending solely to its physical features to observing a conventional use of it (Urqueta Alfaro et al., 2018).

The video data were coded to determine if the joint engagement was coordinated using a novel coding protocol tailored to children with VI (available upon request via e-mail to the first author). This protocol was created based on a review of the literature on CJE, observation of this study's video data, and discussion among the first author and research assistants. The coder first answered the question "does the infant demonstrate an awareness that s/he and the social partner are attending to the same focus?" If the answer was "no", the episode was *not* assigned the code CJE. If the answer was "yes", the episode was coded as CJE. Next, the coder determined which of the infant's behaviors indicated that s/he acknowledged the social partner's participation in their shared attention. These behaviors or CJE indices are as follows. (a) *Vocalization*: infant vocalizes toward social partner; the vocalization is related to the joint engagement. (b) *Body orientation*: infant turns body (whole or part) towards social partner in order to attend to him/her. (c) *Touch*: infant touches social partner (excluding when touch is a means to an end unrelated to the shared focus). (d) *Listening*: infant listens to social partner's vocalizations about the object and/or to the sounds resulting from the social partner's manipulation of the object. Listening behavior can be manifested in several ways, for instance the infant: moves head/ear closer to the source of the sound,

Table 1
Infants' ages and visual diagnoses.

Participant number	Age M = 1 y, 6 m, 27 d SD =3 m, 6 d	Visual diagnoses
1	1 y, 0 m, 10 d	Optic Nerve Hypoplasia
2	1 y, 2 m, 26 d	Norrie's; bilateral retinal dysplasia, cataracts, aphakia
3	1 y, 4 m, 11 d	Ocular Albinism, Left eye Retinal Folds
4	1 y, 5 m, 1 d	Oculocutaneous Albinism
5	1 y, 5 m, 9 d	Right eye cataract, Macular scarring
6	1 y, 5 m, 26 d	Optic Nerve Hypoplasia, Septo-Optic Dysplasia
7	1 y, 6 m, 0 d	Infantile Nystagmus Syndrome
8	1 y, 6 m, 3 d	Right eye Prosthesis, Left eye Iris & Retinochoroidal Coloboma
9	1 y, 6 m, 4 d	Oculocutaneous Albinism
10	1 y, 6 m, 7 d	Cortical Visual Impairment
11	1 y, 6 m, 9 d	Optic Nerve Hypoplasia
12	1 y, 6 m, 17 d	Congenital Cataracts, Aphakia, Glaucoma
13	1 y, 6 m, 25 d	Oculocutaneous Albinism
14	1 y, 7 m, 26 d	Oculocutaneous Albinism, Nystagmus
15	1 y, 8 m, 10 d	Cortical Visual Impairment
16	1 y, 8 m, 16 d	Glaucoma, Left eye Aphakia
17	1 y, 9 m, 28 d	Familial Exudative Vitreoretinopathy
18	2 y, 0 m, 5 d	Cat Eye Syndrome
19	2 y, 1 m, 8 d	Retinopathy of prematurity with bilateral retinal detachments, Right eye cataract, Left eye complete corneal opacity
20	2 y, 3 m, 8 d	Familial Exudative Vitreoretinopathy, Aphakia

Note: y = year(s), m = month(s), d = day(s), M = mean, SD = standard deviation.

responds to the content of the vocalization (e.g., after the social partner verbally requests the object, the infant extends the object toward the social partner), lowers face and stops any actions he was engaged in. (e) *Pause motor*: infant interrupts his body movements in order to attend to the social partner. Attention to the social partner could be manifested in several ways, e.g., body orientation, listening, sensing the social partner's actions through touch. (g) *Gaze switch*: infant shifts his gaze from shared focus to social partner (or *vice versa*) at least one time. The infant does not need to make eye contact. (h) *Gaze point*: social partner points to an object and infant's gaze follows the direction of the pointing. (f) *Give object*: infant gives object to social partner. (g) *Other*: behavior not included in the previous indices. Coding instructions did not provide examples of what behaviors could be coded under "other" because this code was meant to give the coder the possibility of identifying new indices not included in the protocol. Please see the Results section for the behaviors that coders classified as "other". For each CJE episode, all observed indices were coded, that is, multiple indices could be coded. Coding was done using Mangold International's INTERACT (http://www.mangold-international.com).

A first coder (the first author) coded all videos (N = 20). For reliability purposes, each of two second coders (research assistants) independently coded 9 videos. In total, 18 videos were double coded. Consensus for the disagreements was reached through discussion. This study reports the results of the coding after coders achieved consensus.

2.2.2. Visual measures

Measurements of the infants' visual acuity (VA) and contrast sensitivity (CS) were obtained using Preferential Looking (PL) techniques and sweep Visual Evoked Potential (VEP) by an optometrist that was part of the research team that collected the data. Each of these measurements were considered separately: VA with PL, VA with VEP, CS with PL, CS with VEP. The PL is a behavioral test based on an infant's innate preference to look at complex visual patterns (Dobson, 1994; Teller, 1997). For measuring VA using PL, the infant was presented simultaneously with a 5 cm by 5 cm field comprised of black-and-white stripes (gratings) and a 5 by 5 cm homogeneous gray field (a gray target of equal space average luminance). The examiner, who was unaware of the grating's location (to the left or right of a viewing peephole), determined the gratings' location based on the infant's looking behavior. An infant's VA level was determined by the smallest width stripe whose location was correctly determined by the examiner (Dobson, 1994). For measuring CS, the child was presented with a printed schematic "smiley" face paired with a blank page. The contrast between the face and its background was reduced in nine steps over the range from 80 % to 1.6 % Michelson to determine the CS threshold (Haegerstrom-Portnoy, 1993).The VEP involves recording the small electrical signals that are generated within the occipital lobe of the brain while the child views visual patterns (Norcia, 1994). The VEP which does not require a verbal/motor response, may be a more accurate measure of the occipital response to visual stimuli. However, the PL which does require a behavioral response, can be a more accurate estimate of a child's functioning in the natural environment (Watson, Orel-Bixler, & Haegerstrom-Portnoy, 2009).

2.3. Data analysis

2.3.1. Reliability of coders

To assess inter-coder reliability Cohen's kappa (k) was calculated using Mangold International's INTERACT. The obtained k values for the code CJE were 0.98 and 0.90 (for each of the two second coders) suggesting good reliability (a value of 1 indicates perfect agreement). The k values for the nine CJE indices ranged between 0.80 and 0.92 for one second coder. For the other second coder, all but one of the k values were between 0.60 and 0.75 (Table 2).

2.3.2. Participation in CJE

Based on the coding of JE conducted in a prior study, the number of episodes infants participated in JE had a mean of 38.6 (SD = 13.2). In this study, for each child, the percentage of episodes of joint engagement that were coded as CJE were calculated for each infant (hereafter "CJE %"). The percentage of CJE episodes in which each CJE index was present was calculated, as well as the percentage of each behavior classified under the index "Other" (e.g., smiling to caregiver).

Table 2
Inter-rater reliability results measured with Cohen's kappa.

Code	Cohen's kappa	
	Second coder A N = 9 videos	Second coder B N = 9 videos
Coordinated Joint Engagement	0.98	0.90
Vocalization	0.86	0.68
Body orientation	0.90	0.69
Touch	0.88	0.50
Listening	0.86	0.67
Pause motor	0.80	0.60
Gaze switch	0.92	0.70
Gaze pointing	0.92	0.75
Giving object	0.91	0.74
Other	0.89	0.66

Note: Second coder A = Cohen's kappa value obtained by the first coder and second coder A. Second coder B = Cohen's kappa value obtained by the first coder and second coder B.

2.3.3. Relationship between CJE and visual function

To determine if there was a significant relationship between the infants' visual function and their participation in CJE, regression analysis was calculated with infants' age, VA, and CS as independent variables and CJE % as the dependent variable. To determine which independent variables to include in the model and in which order to enter them, correlations were calculated between CJE% and each of the predictor variables (age, VA, and CS), and between all the visual measures (VA (PL), VA (VEP), CS (PL), CS (VEP)). These correlations used Spearman's rho because variables were not normally distributed based on QQ-plots and Kolmogorov-Smirnov test. VA is expressed in terms of the log of the minimum angle of resolution or logMAR. Each 0.1 in logMAR corresponds to one line on an eye chart. Expressing VA in logMAR is a standard convention used because it allows to give equal credit (0.02 log units per optotype) for each letter read, and it linearizes data for the purpose of statistical analysis (Holladay, 1997). Since VA norms vary depending on age, each child's reduction in VA from the age norm was calculated (in logMAR). CS values were expressed as the lowest detectable contrast in percentages. For the VA and CS measures, the higher the value (in logMAR and percentage, respectively) the more impaired the infant's vision. Infants with visual impairment may have deficits in VA and/or CS.

3. Results

3.1. Coordinated joint engagement

The mean number of CJE episodes in which infants participated was 8.9 (SD = 5.9). The CJE % (i.e., percentage of joint engagement episodes coded as CJE) ranged between 0% and 52.6 %, with a mean of 23.5 % (SD = 16.7 %). Fig. 1 shows the CJE % for each participant, only one infant did not demonstrate CJE. Age was not significantly correlated with CJE % (see section 3.2).

Table 3 shows the percentage of CJE episodes in which each CJE index was observed. Indices are shown in decreasing order starting with the index observed in the larger percentage of CJE episodes.

With respect to the infant behaviors coded with the index *other*, the most frequently observed was smiling to the caregiver (50 % of the CJE episodes in which *other* was coded). The remaining behaviors were: pointing/reaching toward object (15 %), expressing emotions toward the caregiver (8.8 %), making gestures (7.5 %), holding hands up seemingly for requesting object (6.3 %), and imitating the caregiver (3.8 %).

3.2. Relationship between CJE and visual function

Considering infants' results with PL and VEP measures, nineteen infants had VA results below age norm (with either or both measures). These infants' reduction in VA from age norm ranged from equal or less than 0.1 logMAR reduction to more than 1 logMAR reduction. One infant did not show VA below age norm. Regarding CS, the lowest contrast detected by the infants ranged between 0.1

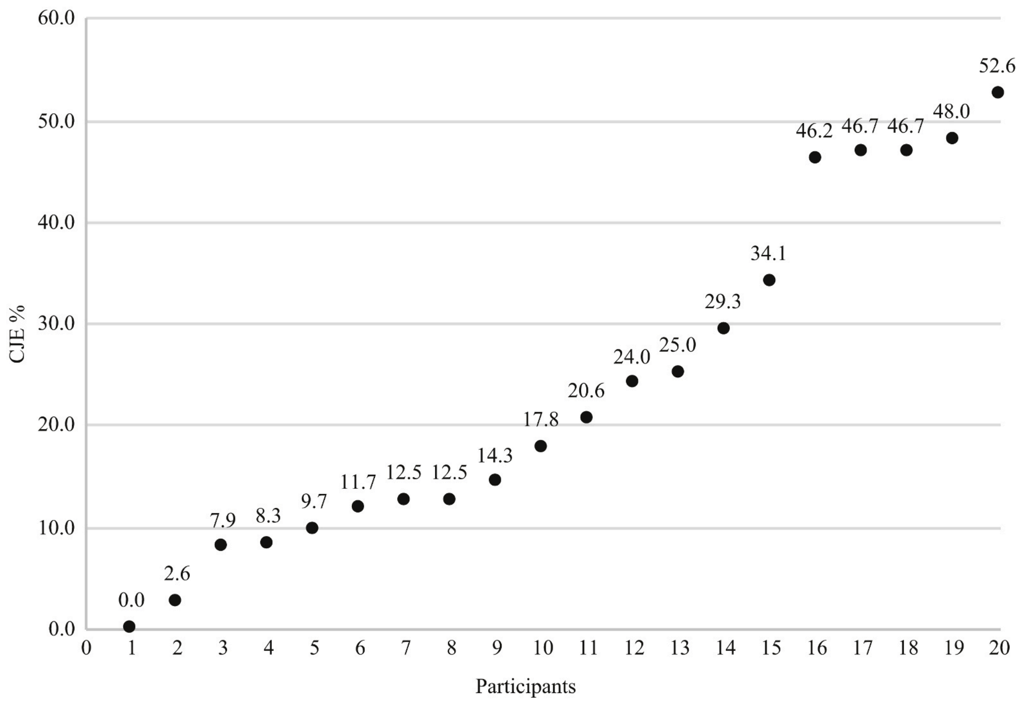

Fig. 1. Percentage of joint engagement episodes coded as coordinated per participant.
Note: CJE % = percentage of joint engagement episodes coded as coordinated.

Table 3
Average percentage of CJE episodes in which each CJE index was observed.

Index of Coordinated Joint Engagement	Mean (SD) %	Minimum-Maximum %
Body orientation	61.8 (29.1)	0.0−100
Gaze switch	54.9 (33.4)	0.0−100
Vocalization	43.1 (28.0)	0.0−100
Other	42.2 (30.0)	0.0−100
Pause motor	39.8 (20.3)	0.0–83.3
Give object	28.3 (22.1)	0.0–62.5
Touch	25.9 (31.3)	0.0−100
Listening	21.7 (27.7)	0.0−100
Gaze point	1.0 (4.5)	0.0–20.0

Note: CJE = coordinated joint engagement, SD = standard deviation.
Minimum: the lowest percentage of CJE episodes in which a given index was observed in the sample. 0.0 % means that the index was not observed in at least one infant.
Maximum: the highest percentage of time a given index was observed in the sample, e.g., 100 % means that the index was observed in all episodes of CJE in at least one infant.

% and 80 %. The CS of sixteen infants were below norm (with either or both measures). The remaining 4 infants did not show CS below age norm. Figs. 2–5 show the results for each infant as well as mean and SD for the sample with respect to reduction in VA from age norm and lowest contracts detected, separately for each measure.

Age was not significantly correlated with CJE %, VA or CS (all p-values > 0.06), and was thus not included in the regression model (Table 4). Based on the Kolmogorov-Smirnov test, the variables CS with VEP and CS with PL were not normally distributed (all p-values <0.023), and CS%, VA with PL, and VA with VEP were normally distributed (all p-values > 0.064).

The two measures (VEP and PL) of VA were significantly positively correlated. This was also the case for the two measures of CS. Likewise, the measures of VA and CS were significantly positively correlated (Table 4). These results suggested that including two or more visual measures in the model could yield to multicollinearity. Because of this and the study's statistical power resulting from a small sample, we included only a single predictor in the model of CJE.

Two visual measures were significantly negatively correlated with CJE %: VA with PL ($r = -0.53$, $p = 0.01$) and CS with VEP ($r = -0.46$, $p = 0.04$) suggesting that the more impaired the child's visual function, the less s/he participated in CJE. We included VA with PL as the single predictor in the model because it was the visual measure with the highest significant correlation with CJE%. This model was significant ($p = 0.01$) and explained 26.8 % of the variance (Table 5).

Keeping in mind that our statistical power may not support adding a second predictor variable to the model, we explored whether adding CS to the model with VEP increased the variance explained. This was not the case (R^2 change = .00) and partial correlations suggested that almost all of the variance was explained by VA with PL ($r = -0.41$), with little unique variance explained by CS with VEP ($r = 0.01$). We also run a regression model entering first CS (VEP) and then VA (PL). Based on the partial correlations obtained, CS (VEP) explained virtually none of the variance ($r = 0.01$), while VA(PL) explained most of it (r = −0.41). Therefore, whether we entered VA (PL) or CS (VEP) first on the model, VA (PL) explained more variance in CJE ($r = -0.41$) than CS (VEP) ($r = 0.01$).

3.3. Exploratory analyses

Given that CJE was measured by using the child's own toys (Toys 1) and a standard set of toys (Toys 2), we explored whether the relationship between the visual measures and CJE % was the same for the two set of toys. We calculated CJE % separately for each set of toys (hereafter "CJE % Toys 1" and "CJE % Toys 2"). The mean of CJE % Toys 2 (22.3 %) was smaller than that of CJE % Toys 1 (29.94 %), but this difference did not reach significance, $t(19) = 1.26$, $p = 0.22$. Based on Spearman's rho correlations, three visual measures were significantly negatively correlated with CJE % Toys 1. VA with PL had the highest correlation ($r = -0.62$, $p = 0.003$),

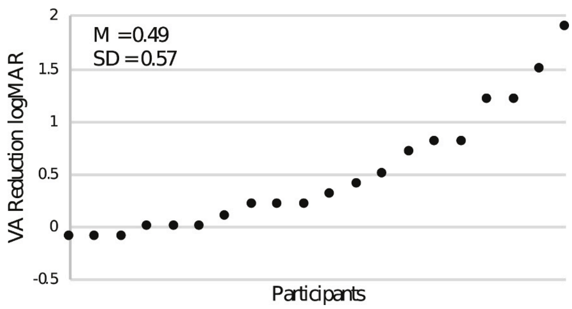

Fig. 2. Infants' reduction in visual acuity from age norm as measured with Preferential Looking techniques.
Note: VA = visual acuity; logMAR = log of the minimum angle of resolution; M = mean; SD = standard deviation.

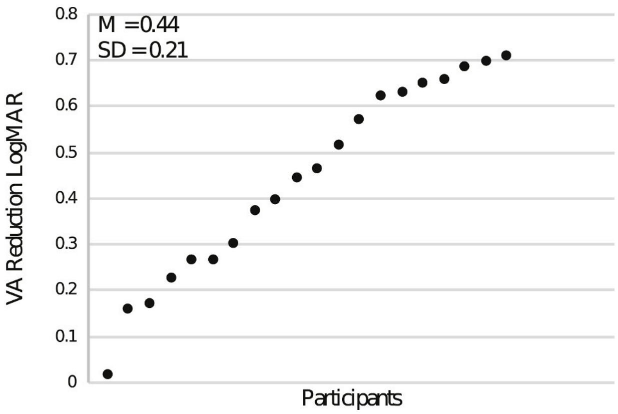

Fig. 3. Infants' reduction in visual acuity from age norm as measured with Visual Evoked Potential.
Note: VA = visual acuity; logMAR = log of the minimum angle of resolution; M = mean; SD = standard deviation.

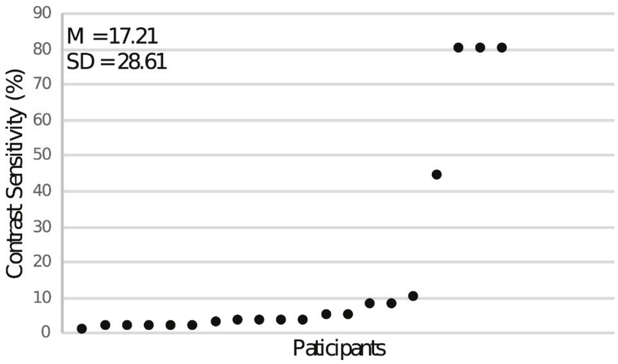

Fig. 4. Infants' lowest contrast detected (Contrast Sensitivity) as measured with Preferential Looking techniques.
Note: M = mean; SD = standard deviation.

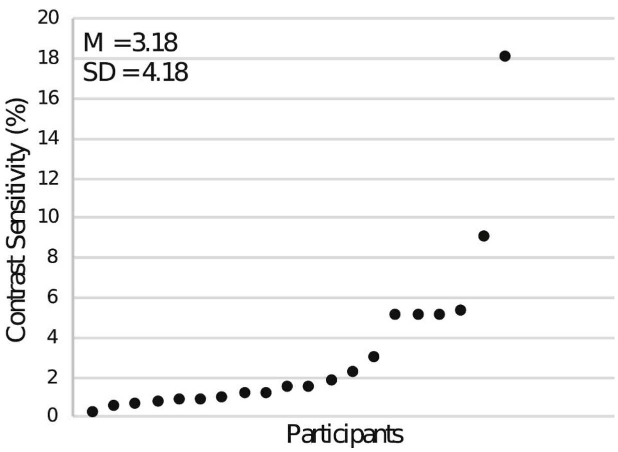

Fig. 5. Infants' lowest contrast detected (Contrast Sensitivity) as measured with Visual Evoked Potential.
Note: M = mean; SD = standard deviation.

followed by VA with VEP ($r = -0.60$, $p = 0.005$), and then CS with PL (r = −0.46, p = 0.03). Similar to the regression modeling of CJE % (combining Toys 1 and Toys 2), the model that explained the most variance in CJE % Toys 1 included VA with PL as single predictor ($R^2 = 0.34$, $p = 0.07$). Adding CS with PL as a predictor did not explain any additional variance (R^2 change = .00) and partial correlations indicated that almost all of the variance was explained by VA with PL ($r = -0.37$; CS with PL: $r = -.004$).

In contrast, none of the correlations between CJE % Toys 2 and the visual measures were significant. With the exception of CS with

Table 4
Spearman's rho correlations (two-tailed) among age, CJE % and visual measures.

Variable	1	2	3	4	5
1. CJE	–				
2. VA (PL)	−0.531*	–			
3. VA (VEP)	−0.433	0.850***	–		
4. CS (PL)	−0.288	0.622**	0.755***	–	
5. CS (VEP)	−0.461*	0.638**	0.626**	0.626**	–
6. Age	−0.091	0.025	−0.148	−0.422	−0.103

Note: CJE = percentage of joint engagement episodes coded as coordinated, VA = visual acuity, PL = preferential looking, VEP = visual evoked potential, CS = contrast sensitivity.
* $p < .05$.
** $p < .01$.
*** $p < .001$ level.

Table 5
Model of Coordinated Joint Engagement.

Predictor	β	*SE*	b	*t*	*p*	R^2	*Adjusted* R^2	*F*	*p*
Intercept	30.75	4.333		7.097	< .001	0.268	0.228	6.605	0.019
VA with PL	−14.90	5.798	−0.518	−2.570	0.019				

Note: VA = visual acuity, PL = Preferential Looking.

PL, all visual measures had negative correlations. As was the case with CJE Toys 1, VA with PL had the highest correlation with CJE Toys 2 ($r = -0.22$, $p = 0.34$). As expected by the correlation results, regression models that included each visual measure as a single predictor of CJE % Toys 2 were all not significant (R^2 ranged between 0.009 and 0.045, *p*-values ranged between 0.36 and 0.76).

4. Discussion

The current study arose from the need to expand the research of CJE in children with VI who are an understudied population that faces challenges in the developmental of this critical developmental milestone. We set to develop a way to identify CJE that is tailored to children with VI, as well as to investigate how impairments in VA and CS impact CJE.

Using a novel coding protocol that did not require normal vision in the child, this study's coders identified CJE in infants with VI with good inter-coder reliability. This result supports the use of this protocol in future studies that aim to measure CJE in infants with VI. The protocol can also be useful for practitioners who need to assess CJE. There is potential for using this protocol with children with ASD whose difficulties with socially directed gaze make them less likely to demonstrate CJE through gaze switch (Bhat, Galloway, & Landa, 2010). Future research could use an adapted form of this protocol to investigate how infants who are blind (who were not included in our sample) demonstrate CJE.

Compared to prior protocols that used one or two behaviors to index CJE, a unique aspect of our protocol is the inclusion of more indices. Our results show that on average, eight of the nine indices were observed in roughly 22 % or more of CJE episodes. The exception was the index *Gaze point* (1.0 %) which may be a consequence of this behavior's visual complexity (i.e., detecting the pointing gesture of the social partner and locating its reference in space). Indeed research indicates that children with VI have difficulties understanding and using pointing (Pérez-Pereira & Conti-Ramsden, 2005). If we consider the indices' prevalence per infant, six out of nine indices were observed in between 0% and 100 % of CJE episodes. This wide range of results highlights the variability of how CJE can be identified, with some infants never showing a given index while others demonstrating it all the times they participated in CJE. Results suggest that if some indices were eliminated (i.e., those that were less prevalent in the sample), the protocol would be less sensitive which would increase the risk of under detecting CJE in children with VI.

By far, the most observed CJE index in the sample was *Body orientation*. This finding is in line with evidence that children with VI's focus of attention can be determined by their body orientation (Preisler, 1991). Interestingly, the second most prevalent index was *Gaze switch*, with the caveat that our protocol did not require eye-to-eye contact with the caregiver. This result in a sample of VI infants all of whom had residual vision is particularly relevant when we consider that the great majority of children with VI have residual vision and only a small minority are totally blind (Lueck & Heinze, 2004). It is also worth noticing that the index *Other* had the fourth highest prevalence. Smiling and otherwise expressing emotions to the caregiver constituted the majority of the behaviors coded as *Other* (e.g., expressing frustration to the caregiver when the child cannot perform an action with the object like activating a toy). This result is in line with studies that show how infants are more likely to emote to their caregivers during CJE than outside of it (Adamson & Bakeman, 1985; Mundy, Kasari, & Sigman, 1992). Based on our findings regarding the index *Other*, we propose that the following two indices could be added to the protocol. One that encompasses the infants' emotional expressions to the caregiver (including smiling), and another for the child's gesturing to the caregiver (e.g., pointing/reaching toward the object as a mean of requesting the caregiver to give the object to the child). Still, we would keep the index *Other* so that coders can identify behaviors that are idiosyncratic to specific children.

Prior research indicates that the amount of residual vision influences the developmental outcomes of children with VI in that those with the most severe levels of VI show the largest lags in achieving milestones (Dale & Salt, 2007). In line with this evidence, this study's correlation results suggest that the more impaired infants' VA and CS were, the less they showed CJE. Contrary to our expectation, the regression analysis indicates that VA (measured with PL) explained more variance in CJE than CS. A prior study in infants with VI (using the same sample and video data analyzed in our study) found that when modeling their participation in joint engagement *without* differentiating between coordinated and supported, CS (measured with PL) was the best predictor (Urqueta Alfaro et al., 2018). Taken together, these findings suggest that although impairment in CS is a better predictor of children with VI's total capacity for participating in joint engagement (whether or not they acknowledge the social partner's participation in the shared attention), impairment in VA is more relevant for predicting these children's ability to *acknowledge* the social partner's participation in the joint engagement. More research is needed to investigate why VA rather than CS was the stronger predictor of CJE. One possibility is that CS is more important for *supported* joint engagement because in this type of joint engagement the child's capacity to share attention is more dependent on the social partner's scaffolding (i.e., actively prompting the child to share attention). This scaffolding may involve the social partner's facial expressions. Perception of facial expressions is more dependent on CS than VA. This could explain the prior study's finding of CS being the best predictor of joint engagement episodes that, unlike the present study which only focused on CJE, also included supported joint engagement. Compared to supported joint engagement, CJE requires a more active role in the child, which includes the child utilizing fine motor skills to manipulate the object. There is evidence that lower visual acuity is related to poorer fine motor skills (O'Connor, Birch, Anderson, & Draper, 2019). This may be a factor contributing to our finding that VA explained more variance in CJE, as children with worse VA would have more difficulty manipulating objects which could restrict their ability to participate in CJE. It is noteworthy that in these two studies the variables (whether CS or VA) that explained the most variance were measured with PL rather than VEP. This suggests that the impact of VI in children's participation in joint engagement depends more on the child's capacity to integrate visual information in their behavioral responses (information provided by PL), than on the occipital response to visual stimuli (data gathered by VEP).

In an exploratory regression analysis we found that if we considered only CJE episodes in which children used their own toys (Toys 1), VA with PL was again the best predictor. However, when we modeled only CJE using the standard set of toys (Toys 2), no visual measure was a significant predictor. We did not intend to investigate the impact of type of toy and thus did not counterbalance their presentation. Consequently, further research is needed to disentangle how fatigue (suggested by the lower CJE % observed with Toys 2) versus type of toy may help explain these diverse results. It is possible that infants experienced visual fatigue (Lueck, Chen, Kekelis, & Hartmann, 2008), utilized less visual input during the last 15 min of the observation, and consequently visual measures were not significantly related to participation in CJE when they used Toys 2. Future research on the effect of type of toy can inform the assessment of CJE in children with VI. Whereas intervention practices promote the use of familiar toys when the goal is to observe these children's higher functional levels (Lueck et al., 2008), many assessments of CJE use toys that are novel to the child. Future research should investigate whether children with VI, as was the case in our study, do show more CJE when using familiar toys.

Keeping in mind that our sample size limits the generalizability of our findings to the population of children with VI, our results contribute to understanding the normative development of CJE in these children. We found that infants with VI, whose parents received developmental counseling, participated in CJE at ages between roughly 1 and 2 years. Still, one infant did not show CJE which points to individual differences in CJE development. We did not identify factors that could contribute to the lack of CJE in this infant because other children in the sample, who had similar demographic and VI characteristics, did show CJE.

Although we are of the opinion that to determine abnormal development in a child with VI s/he should be assessed using norms based on children with VI, we do recognize that many researchers determine this instead based on the norms of sighted children. Therefore, a question that may arise is how our results compare to those of sighted children. The answer is not straightforward because prior studies used different CJE indices and testing methodologies (i.e., parental questionnaire, structured play). For instance, Bakeman and Adamson (1984) also used free play but a different metric of CJE (i.e., percentage of time in CJE) than in the present study (i.e., percentage of episodes of joint engagement that were coordinated). In their study, sighted infants—aged between 12 and 18 months—spent between 3.6 % and 26.6 % of the time in CJE on average. In the present study, 23.5 % of joint engagement episodes were coordinated in infants with VI—aged between 12 and 24 months. These results cannot be directly compared however given that the studies differ on how CJE was quantified.

Our results should be considered in the context of study limitations. As previously indicated, the sample size limits the generalizability of our findings. It also limits the statistical power and consequently our regression analyses were restricted in the number of predictor variables entered in the models. Unlike prior studies that used structured play contexts we measured CJE during free play and thus did not instruct the caregiver to prompt CJE. This may have led to an underestimation of CJE as it is possible that if infants had been prompted they would have shown more CJE. Two characteristics of the sample should be kept in mind. First, all caregivers received developmental counseling and thus our findings cannot be extrapolated to children whose caregivers did not have such support. It is possible that without developmental counseling children with VI and their caregivers would show less CJE. Second, all of the infants in the present study had residual vision and our coding protocol included two indexes that required some level of vision (i.e., gaze switch and gaze point). Therefore, our findings cannot be extended to the population of children who are blind.

4.1. Conclusions

In summary, this study demonstrates that CJE can be identified in children with VI using behaviors that do not require normal vision, with good inter-coder reliability. Some of the most commonly observed behaviors were body orientation, vocalization, and smiling toward the caregiver. Gaze switch was also prevalent suggesting that this behavior can be used to index CJE in children with VI that have enough residual vision. Results also show that VA, measured with PL, explained the most variance in CJE. This suggests that the lower a child's capability to behaviorally respond to detailed visual information, the lower his/her participation in CJE. These findings highlight the need for intervention to promote the development of CJE in children with VI.

Policy and ethics statement

The authors declare that this research was conducted in accordance with APA ethical standards in the treatment of human subjects included in the study sample.

Data availability statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

Declaration of Competing Interest

The authors report no declarations of interest.

Acknowledgements

Gratitude is expressed to the Blind Babies Foundation, CA; and the infants and caregivers who participated in this study. We would also like to acknowledge our research assistants Christine Xu and Thérèse Pham for their exceptional work and dedication. Urqueta Alfaro was supported by a post-doctoral fellowship by the Institut Nazareth et Louis-Braille du CISSS de la Montérégie-Centre, Montréal. Wittich is supported by a Junior 2 chercheur boursier Career Award, funded by the Fonds de recherche du Québec - Santé (#281454).

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