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Joint engagement in infants and its relationship to their visual impairment measurements

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Sadly, our beloved colleague and co-author Valerie Morash passed away in January 2017. She contributed greatly in the statistical analyses in this project and reviewed the manuscript for submission. However, she passed away during the submission of this paper. She was therefore not involved in the correction process.

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ABSTRACT

Coordination of attention between a social partner and an external focus of shared interest, called joint engagement, is associated with positive developmental outcomes such as better language, socio-emotional, and theory of mind skills in sighted infants. Current measures of joint engagement rely on an infant's visual behaviors, making it difficult to study joint engagement in infants with low or no vision. In a naturalistic observational study, 20 infants with various levels of visual impairments – mean ages: 1.08 years (N = 9) and 1.62 years (N = 18), were videotaped during 30-min free play sessions with their caregivers. Seven infants were tested at both ages. Videos were coded to determine the percentage of time the dyads participated in joint engagement. Results showed that all visually impaired infants participated in joint engagement, with a significant increase between earlier and later ages. Infants' visual impairment levels were described in terms of visual acuity and contrast sensitivity as measured using both visual evoked potential and preferential looking techniques. Of the visual measurements, infants' reduction in contrast sensitivity measured with preferential looking, alone, predicted the infants' percentage of time in joint engagement across ages. Contrary to prior research that exclusively focused on visual acuity, this finding supports the need to include contrast sensitivity measurements in studies with visually impaired infants.

1. Introduction

Around the end of the first year of life, sighted infants start to participate in joint engagement (JE), that is, in interactions where they coordinate their attention between a social partner and an external focus (object/event in the environment) that is of interest to both the child and his/her social partner (Bakeman & Adamson, 1984). It is well documented that JE relates to positive developmental outcomes in sighted children such as better language, socio-emotional, and theory of mind skills (Carpenter, Nagell, & Tomasello, 1998). It is important to characterize JE development in all infants so that disturbances are recognized promptly. While JE development has been addressed by numerous studies with sighted infants, only a few studies have addressed this question with visually impaired (VI). The few studies that exist include either very few subjects or second-hand reports, which limits their validity. Research with this population is particularly necessary since VI infants can have difficulties in early interactions such as engaging in contingent and mutually responsive exchanges with caregivers, and engaging with objects, that in sighted infants are deemed germane to JE development (Bigelow, 2003). The current study addresses this research need by reporting 20 VI infants' time expended in JE (using both visual and non-visual senses), and the predictive role of an infant's contrast sensitivity for participation in JE.

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1.1. JE in sighted infants

Developmentally, infants first participate in “supported” joint engagement (SJE). SJE depends on the social partner attracting the attention of infants to his/her actions with objects. Although the child is aware of the caregiver’s action with respect to the object, the child’s attention remains primarily on the object. Later in development, infants participate in “coordinated” joint engagement (CJE). In CJE, infants themselves can initiate the sharing of attention and overtly acknowledge the social partner’s participation in their common engagement (Adamson, Bakeman, Deckner & Nelson, 2012; Bakeman & Adamson, 1984, 1986). In research with sighted infants, indexing of such acknowledgement has heavily relied on the child’s gazing back and forth between social partner and object (Carpenter et al., 1998). This same behavior is utilized to index the concept of “joint attention”. Despite this common index, authors vary in their claims as to what this milestone implies for children’s social cognition. For some joint attention authors (Carpenter et al., 1998), it implies that the child has an initial understanding of others as intentional agents in the sense of having goals and mental representations that inform their actions. For coordinated joint engagement authors, it implies that the child voluntarily and repetitively notices the social partner’s role in their attention to a common focus without claiming the child understands others as intentional agents (Bakeman & Adamson, 1984). By the last half of the second year, symbols are being incorporated within JE (“symbol-infused”), allowing for communication regarding absent or imaginary references (Adamson, Bakeman, & Deckner, 2004). Evidence shows a relationship between JE and healthy development. Sighted infants’ participation in CJE and symbol-infused SJE correlates positively to their receptive and expressive language at later ages (Adamson et al., 2004; Mundy et al., 2007). They show more “functional play” (using objects in conventional ways) during CJE and more stereotypical play outside of it (Bigelow, 2004). Participation in CJE and symbol-infused JE correlates positively with sighted infants’ performance at preschool age in theory of mind tests (Charman et al., 2000; Nelson, Adamson, & Bakeman, 2008). Participation in CJE in infancy positively correlates to emotional regulation skills at both concurrent and later ages (Morales, Mundy, Crowson, Neal, & Delgado, 2005; Vaughan Van Hecke et al., 2012).

1.2. Risk factors in VI infants

Prior to JE, sighted infants participate in contingent and mutually responsive exchanges with caregivers (Adamson & Mc Arthur, 1995). VI infants are at risk of engaging less in such interactions because several behaviors reported in these infants, when interpreted by caregivers as lack of interest, decrease positive social exchanges (Warren, 1984). These behaviors include: responding to social interaction with decreased/absent eye contact, gaze or smiling (Lueck, Chen, Kekelis, & Hartman, 2008); averting gaze and turning head/body away from caregivers; and initiating play interactions with their mothers less often than sighted peers (Cass, 1998; Kekelis & Anderson, 1984; Rogers, 1988; Rogers & Puchalski, 1984). However, it is unclear whether this perceived lack of interest in social interactions is a result of an observer’s biases regarding what indexes an infant’s interest. VI infants demonstrate interest in objects or people by active tactile exploration (Fraiberg, 1977); react to mother’s approach by increasing motor activity (Preisler, 1990); and initiate communication with caregivers as frequently as sighted peers (Pérez-Pereira & Conti-Ramsden, 2005). Prior to JE, sighted infants also experience how their behaviors impact their social partners by seeing how others react to their own facial expressions. Since VI infants have reduced or no access to such visual cues, if their social partners do not use other cues, these infants will have fewer opportunities to perceive their actions as having an effect on others (Bigelow, 2003). However, there is evidence that caregivers can learn to interpret and sensibly respond to VI infants’ signals through non-visual means (Als, Tronick, & Brazelton, 1981; Loots, Devisé, & Sermijn, 2003).

JE requires participants to recognize when others attend to an external focus. VI infants risk engaging less with objects; reaching for objects in sighted peers is observed by 4–6 months of age, whereas reaching upon hearing objects’ sound cues emerges later—10-12 months—in both sighted and visually impaired children (Warren, 1984). A connection between VI infants’ advancements in object permanence and JE was reported in two congenitally blind infants. These infants’ CJE and object permanence were assessed longitudinally. CJE was observed only after infants had started reaching for objects on sound cues (Bigelow, 2003). VI infants can also have less experiences encountering objects in the environment because they may achieve independent locomotion at later ages than sighted peers (Brambring, 2006). When VI infants attend to external foci, caregivers can have difficulties identifying infants’ attentional cues and/or responding in ways within infants’ perceptual capabilities (Bigelow, 2003; Preisler, 1990).

There is evidence that VI infants develop JE more slowly than sighted peers. Social games between VI infants and their caregivers over-rely on repetitive vocal games, to the detriment of games involving objects (Dale & Salt, 2007). In a study of 10 VI children, 1-year-old blind infants had difficulties sharing attention on objects, and even very low vision positively impacted this ability (Preisler, 1990). A study of two congenitally blind infants reported emergence of CJE at later ages than in sighted peers (18 and 23 months versus 12 months, Bigelow, 2003). One-to-three-year-old children who have only light perception or worse visual condition had significantly lower CJE skills scores than sighted peers (Dale, Tadić, & Sonksen, 2013). Compared to sighted peers, VI preschool children scored significantly lower in their ability to establish and maintain CJE (Tadić, Pring, & Dale, 2009).

Two other lines of research indicate the risk of JE development in VI infants. First, atypical JE is part of the developmental regression/stasis reported in a sub-group of blind/severely visually impaired infants (Cass, Sonksen, & McConachie, 1994; Dale, 2005; Dale & Sonksen, 2002). Second, atypical JE in VI infants, compared to their sighted peers’ developmental patterns, is considered by some authors “autistic-like”, and used by clinicians to diagnose autism (Hobson, 2005; Naber et al., 2007; for a contrasting perspective see Pérez-Pereira and Conti-Ramsden, 2005). Normative development of JE in VI infants has not been fully characterized, and is currently assessed by comparing it to what is typical in sighted infants. This is problematic. Comparisons between the population of VI children and that of typically sighted children can shed light on the similarities and divergences in their normative developments. They can also advance our understanding of developmental processes by documenting how pathways different than the ones found in sighted children lead to equivalent developmental outcomes. However, findings on sighted infants cannot be used as the standard against which to determine what is normal or abnormal in a VI infant's development. As Warren (1978) argued, the environment to which VI children adapts in presents them with a set of different challenges than the ones faced by their sighted peers. If VI children take longer to demonstrate JE, it may be due to the different demands this posits in children who rely on haptic and auditory information. Without knowledge of normative JE development in this population, VI infants are at risk of both being misdiagnosed as autistic and missing intervention warranted by true abnormalities.

Research on this topic is also needed to inform current theories of joint engagement, which hardly consider the role of senses other than vision. To index infants' attentional focus, researchers have most often considered infants' direction of gaze to be the least ambiguous indicator. This is particularly evident in studies that index pre-verbal infants' acknowledgement of the shared focus by the infants' gaze switching between a toy and their social partner (Carpenter et al., 1998). By definition, JE can be established using non-visual cues; for instance, when a child feels that his/her hand and an adult's hand are tactually exploring the same object. Although some research acknowledges the limitations of focusing on infants' visual behavior (Adamson & Bakeman, 1985; Adamson et al., 2004; Carpenter et al., 1998), and documents JE episodes in which sighted infants attend to adult's voice but not direction of gaze (Leekman & Wyver, 2005; Rossano, Carpenter, & Tomasello, 2012), most research has focused on "visual" joint engagement. Thus, current theories provide an incomplete explanation and may overemphasize the role of vision.

The current study had two goals. The first goal was to contribute to the characterization of JE development in VI infants. We documented in 20 VI infants (without additional disabilities) the ages at which they demonstrated JE and the percentages of time they participated in JE. VI infants and caregivers were videotaped at home during a 30-min free play session with two sets of toys. Free play was used because we were interested in the amount of time VI infants and their caregivers shared attention when left to their own devices. Since most VI children have residual vision, our purpose was not to focus solely on JE through non-visual means, but rather to report JE irrespective of what senses the VI children used. In sighted infants, participation in JE increases with age (Adamson et al., 2014). To test the hypothesis that JE increases with age in VI infants, we compared percentage of time in JE between the two ages tested: 1.08 (SD = 0.12) and 1.62 (SD = 0.23).

The second goal was to investigate the hypothesis that contrast sensitivity (ability to detect differences in the brightness of or subtle shades of gray in large objects) is a stronger predictor of JE than visual acuity (vision for detail). Unlike previous studies that only reported visual acuity (VA) to determine the extent of infants visual impairment, the current investigation also measured contrast sensitivity (CS). We related infants VA and CS to the amount of time they participated in JE. We chose not to measure only VA because VI children's visual impairment may impact visual ability beyond the effect of visual acuity reduction. We focused on CS because it is a better predictor of visual attention than VA (Orel-Bixler, Haegerstrom-Portnoy, & Dornbush, 1983), and due to its relevance for discriminating objects against backgrounds, navigating the environment (e.g., seeing stairs and curbs), and detecting the faint shadows in faces that carry the visual information related to facial expressions ("Lea Contrast Sensitivity," n. d.; Orel-Bixler, 2014). We hypothesized that CS plays a role in JE because of the relevance of CS to experiences that in sighted children are deemed germane to JE development, specifically: finding objects, moving in space, and attending to others facial expressions.

2. Methods

2.1. Participants

Sample design considered the following. First, VI infants show a wide range of visual acuity, including no light perception (total blindness), low vision (warranting accommodations like braille), and VI (reduced visual acuity requiring large print or > 3x magnification). A minority of VI children are totally blind (Lueck, 2004). The way visual impairment influences development varies greatly depending on residual vision; skills achieved by VI infants at later ages than sighted peers show the greatest lags in those with most severe VI (Dale & Salt, 2007). Therefore, we report on a sample of infants with a range of visual impairment based on visual acuity. Second, children with similar VI levels can develop at different rates depending on their families' access to developmental counseling (Warren, 1984). Thus, our sample includes VI infants that were receiving similar types of counseling. Lastly, research with children who are solely visually impaired is challenging since they are a low incidence population, and in western societies the majority of these children have additional disabilities (Sonksen & Dale, 2002). Understandably, previous studies that included behavioral observation measurements of JE included small samples. Bigelow (2003) reported on 2 congenitally blind infants and Preisler (1990) reported on 7 blind and 3 severely VI infants. We investigated a larger sample than in previous studies and included infants with visual impairment of varying severity.

We collected data from 20 VI infants (10 female, 10 male) with no additional disabilities (per infants' medical records and/or caregiver report). Due to the low incidence of this population, they were recruited either as longitudinal (N = 7) or cross-sectional subjects (N = 13), per caregiver's preference. Six were White, 5 Hispanic, 2 Asian, 1 African-American, and the remaining 6 were of more than one race/ethnicity. Sixteen infants were born full term. Four were born prematurely, mean gestational age of 30.75 weeks (range: 24–34 weeks). Reported ages were corrected for prematurity by subtracting the number of weeks infants were born prematurely from their chronological ages (Emory Department of Pediatrics, n. d.). All but two of the infants were receiving in-home intervention, ranging from one to two 60-to-90-min monthly visits by specialists in VI children. The other two infants were being counseled on a less regular basis. Eighteen caregivers were the biological mothers, one was the adoptive mother, and one was the biological father.

The University of California, Berkeley Committee for the Protection of Human Subjects approved the study's protocol. Infants

Table 1
Subjects' ages and visual diagnoses.

Subject Number	Age 1 N = 9 M = 1.08 SD = 0.12 Age Range = [0.91, 1.24]	Age 2 N = 18 M = 1.62 SD = 0.23 Age Range = [1.36, 2.27]	Diagnosis
1	1.15	1.57	Oculocutaneous Albinism
2	0.91	1.42	Oculocutaneous Albinism
3	—	2.27	Familial Exudative Vitreoretinopathy, Aphakia
4	1.03	1.49	Optic Nerve Hypoplasia, Septo-Optic Dysplasia
5	1.24	1.53	Optic Nerve Hypoplasia
6	1.09	1.51	Oculocutaneous Albinism
7	—	1.52	Cortical Visual Impairment
8	1.00	1.5	Infantile Nystagmus Syndrome
9	—	1.44	Right eye cataract, Macular scarring
10	1.24	—	Norrie's; bilateral retinal dysplasia, cataracts, aphakia
11	0.98	1.51	Right eye Prosthesis, Left eye Iris & Retinochoroidal Coloboma
12	—	1.8	Retinopathy of prematurity with bilateral retinal detachments, Right eye cataract, Left eye complete corneal opacity
13	—	2.01	Cat Eye Syndrome
14	—	1.66	Oculocutaneous Albinism, Nystagmus
15	—	1.83	Familial Exudative Vitreoretinopathy
16	—	1.55	Congenital Cataracts, Aphakia, Glaucoma
17	1.03	—	Optic Nerve Hypoplasia
18	—	1.56	Cortical Visual Impairment, Hydrocaphalus
19	—	1.71	Glaucoma, Left eye Aphakia
20	—	1.36	Ocular Albinism, Left eye Retinal Folds

—: Indicates child was not part of corresponding age group.

were recruited through the collaboration of the Blind Babies Foundation, a non-profit that provides developmental services for VI infants, and the patient population in the Infant/Toddler Clinic and the Special Visual Assessment Clinic at the UC Berkeley School of Optometry. Prior to study procedures, informed consent was obtained from infants' caregivers.

2.2. Ages observed

Infants were videotaped at home interacting with their caregivers. Recordings were scheduled as closely as possible to infants' ages of 12 and 18 months. These ages were chosen because research with sighted infants shows that JE emerges toward age 12-month and it becomes more prevalent by 18-month (Adamson et al., 2014). The collected data was grouped into two ages by dividing the sample at the midpoint between ages 12 and 18 months (1 year, 3 months). This resulted in two age groups with means of 12 and 19 months of age. Age 1 infants were younger than 1 year, 3 months. Nine infants (6 females, 3 males) were observed at this age, mean fractional age of 1.08 (SD = 0.12). Age 2 infants were older than 1 year, 3 months. Eighteen infants (10 females, 8 males) were observed at this age, mean fractional age of 1.62 (SD = 0.23). Of these, 7 infants (6 females, 1 male) were also observed at age 1. Table 1 lists infants' ages and VI etiology. Fig. 1 shows the age distribution.

2.3. Procedures

2.3.1. Observational procedure

Thirty-minute video recordings were performed at each infant's home, in a room chosen by the caregiver. Two wide-angle-lens cameras were placed in front of and behind the dyad. Twenty-seven videos were collected, mean duration of 29.9 min (SD = 6.03 s).

2.3.2. Observational conditions

Caregivers were instructed to play with their infants as they would normally do. Since VI children perform at their best with familiar objects (Lueck et al., 2008), caregivers' were asked to use their infants' toys. To counteract differences in toy quality and quantity based on the child's socioeconomic level, caregivers were also asked to use a standard set of toys (Table 2). We were not interested on the effect of the different sets of toys, so we did not counterbalance the presentation of the toys (child's and standard). Our concern was to present toys in a sequence that would more likely keep infants engaged. We reasoned that an infant would be more likely to be engaged in the second 15-min session if we presented novel toys rather than familiar toys. Thus, the order of presentation was first the child's toys, then the standard set. During the videotaping, the investigator waited in a different room.

2.3.3. Video data coding

This study coded each second of time into two mutually exclusive and exhaustive codes: JE, when the child is concurrently engaged with both an object and a person; and Non-JE, when the child is engaged solely with an object, solely with a person, or solely

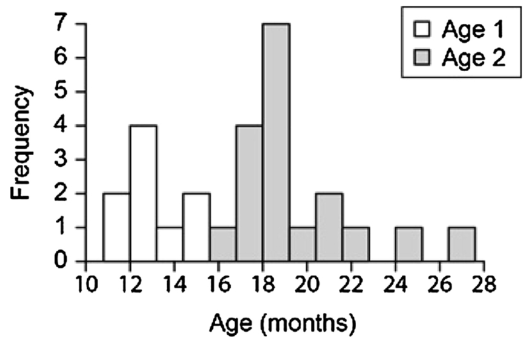

Fig. 1. Subjects' age distribution.

with his/her own body. First, coders had to decide when a child was in an engagement state. An infant was considered to be in state of engagement given either of two indicators: a. The infant was attending to the object and/or person using any one or a combination of senses. Most VI infants in our sample had residual vision, thus attention was indexed by behaviors that could use only the visual sense, only non-visual senses, or a combination of them, for instance: turning body in the direction of speaker, exploring a toy with the hands, gazing and/or reaching toward person or object. b. The infant was performing an action that involved the object and/or person. Second, coders had to decide if the engagement state was JE or non-JE. Definitions of JE and Non-JE codes are based on the protocol developed by Bakeman and Adamson (1984; Adamson et al., 2000). Some adaptations (described below) were done based on preliminary review of video data collected from the sample at ages not reported in this study.

2.3.4. Code: JE

Unlike Adamson and Bakeman's protocol (1984), this study did not differentiate between supported and coordinated JE. We considered that, before tackling the task of indexing CJE without relying on infants' visual behavior, we had to first determine if a basic coding decision of whether the VI child was in JE or not could be realized. In this study, the infant was coded in a state of JE if all three of the following conditions were met:

- The infant was engaged with the same object/event that the caregiver was engaged with. As explained above, 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.
- 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, JE was not coded.
- 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 cultural use of it.

There are two other differences between our study's protocol and that of Bakeman and Adamson's. One, if the caregiver called the child's attention to an object and as a consequence the child attended to it, the child was considered to be in JE, even if the child did

Table 2
Standard set of toys.

Toy with 5 fine-motor operated pop-up figures
Three mittens with different textures
Red mylar pom pom
Four different rattles
Two books with drawings/pictures and textures
Three bells of different color and sound
Radio toy
Metallic bowl
Puffy ball (elastic)
Rolling red car
Six single colored cubes (wood)
Four single colored cylinders (resin)
Cube with textures
Eight big Lego pieces
Shaker
Light-up toy

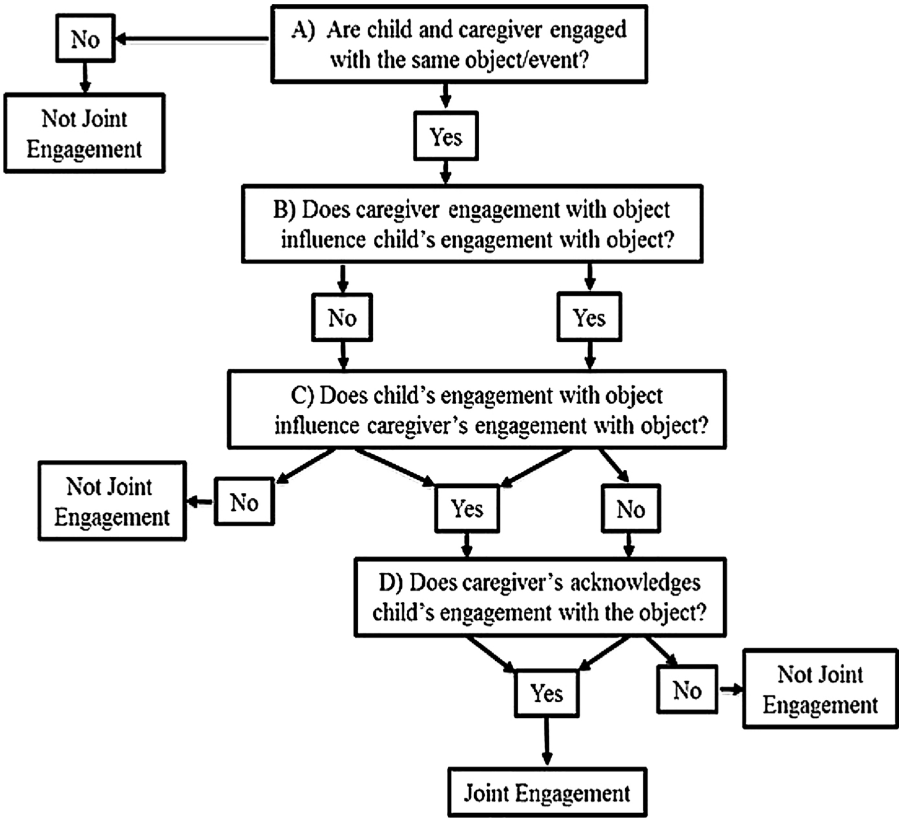

Fig. 2. Coding decision flow chart.

not eventually manipulate the object. In the original protocol, this situation could be coded instead as "onlooking", a type of non-JE. Two, in their protocol, an engagement state must last at least 3 s. However, our preliminary video review showed that some of the infants participated in JE episodes lasting less than 3 s. It was decided that a more accurate report would include such episodes. Thus, the code JE did not have to last at least 3 s.

2.3.5. Code: non-JE

Following Bakeman and Adamson's protocol (1984), the infant was considered to be in a state of Non-JE if the infant was in any of the following situations: engaged only with a person or an object; searching for something to engage with without attending to an external focus for at least 3 s; crying without attending to any external focus; and observing the caregiver acting on an object when the caregiver had not called the child's attention to the activity. Additionally, we coded Non-JE when the child was engaged with movement of his/her whole body or part of it (without involving an object). It was considered that the child was not paying attention if he/she did not direct any of his/her senses to the external focus (e.g., body is not oriented toward focus, child is not listening to sounds from focus, child is neither gazing at nor touching the focus).

Fig. 2 shows a coding decision flow chart.

2.3.6. Coding procedures

Videos were coded in their entirety, in an exhaustive and exclusive manner, that is, each second was coded as only one of the two codes (Bakeman & Quera, 2011). Behaviors were coded as timed events since states of engagement extend over time and thus their extension was deemed more relevant than their counts (Yoder & Symons, 2010). Coders viewed a video until they considered that a change in the child's engagement state had occurred (from JE to Non-JE, or vice versa). Then coders reviewed the relevant portion of the video as many times as needed to determine the transition between states. Codes were entered automatically into computer files using Mangold International's INTERACT (http://www.mangold-international.com).

A first coder—the first author—coded all videos ($N = 27$). For reliability purposes, a second coder—the third author—coded 48.15% of the data ($n = 13$). To approximate the study's full sample age distribution, more videos in the reliability sample corresponded to infants observed at age 2 ($n = 9$), and fewer videos at age 1 ($n = 4$). After both coders independently coded a video, Cohen's kappa and percentage agreements were calculated. Because there is no value of kappa that is universally acceptable, we followed the social sciences literature and set this study's reliability criterion as an agreement of 80–85% (Bakeman & Quera, 2011). If the percentage agreement decreased noticeably between two coded videos, coders reviewed their disagreements before coding a new case (coders did not code again the same video). Coders were masked to infants' visual measurements.

2.3.7. Vision examination

Infants underwent a vision examination in the Clinical Research Center at the University of California at Berkeley School of Optometry (by the fourth author). Measurements of visual acuity (VA) and contrast sensitivity (CS) were obtained using Preferential Looking techniques (PL) and sweep Visual Evoked Potential (VEP). 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 (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), with both fields printed on a 50 cm by 25 cm grey card. 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. Six cards enabled the presentation of decreasing stripe widths representing finer visual acuity. An infant's VA level was determined by the smallest width stripe whose location was correctly determined by the examiner using the acuity card procedure described by Dobson (1994). Using a similar protocol, CS was measured using PL. 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 infant views pattern-reversing stripes on a video monitor located at 50 cm. To record the signals, 5 sensors (3 active, 1 reference and 1 ground) were placed on the infant's scalp over the occipital lobe of the brain. Infants looked at the video monitor for each 10-s trial during which 19 different black and white stripe widths were presented. The amplitude of the VEP response decreases proportionately as the width of the stripes reduces across the trial. A computer analyzed the VEP response and extrapolated the VEP amplitude versus stripe width function to zero amplitude to report the infant's VA. Similarly, the contrast sensitivity threshold was measured by altering the contrast of a 0.5 cycle/degree grating over the range of 1–80% Michelson contrast, and extrapolating the VEP amplitude versus contrast function to zero amplitude (Norcia, 1994). We used these measurements because they each provide different information. Whereas the VEP tells us about the integrity of a child's neural substrate for visual processing, the PL indicates the level of vision such child can access. Behavioral based tests like PL may be affected by the child's visual impairment and performance ability. An electrophysiological technique like 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 can be a more accurate estimate of a child's functioning in the natural environment (Watson, Orel-Bixler, & Haegerstrom-Portnoy, 2010).

2.4. Data analysis

2.4.1. Reliability of coders

To measure reliability, Cohen's kappa was calculated using the GSEQ program (Bakeman & Quera, 2011). Events were aligned in terms of a time window composed of the same number of seconds for each coder. As in previous research (Adamson et al., 2014), a tolerance of 2 s was allowed, which lead to comparing 5-s windows—the current second plus 2 before and after it. The comparison considered each second for coder 1 and looked for a match in any time unit of the 5-s window from coder 2. If coder 2 assigned the same code as coder 1 in any of those 5 s this was counted as an agreement (Bakeman, Quera, & Gnisci, 2009).

Since there is no universally accepted gold standard to evaluate kappa values, two other statistics were calculated to assess reliability. One, for perfect agreement to be possible (a Kappa value of 1), coders must attribute codes similarly. If coders favor codes differently, the Kappa value cannot be 1. Thus, the maximum value of Kappa was calculated to determine what would be the value when coders in this study agreed to the maximum extent possible given their biases (Bakeman & Quera, 2011). The coders' biases are described by the bias index, a numerical value that expresses the extent the coders disagree on the proportion they determine a code as present or absent (Sim & Wright, 2005). Two, because we do not know the real state of affairs in the world, coders' accuracy is estimated using the agreement between two fallible coders (Bruckner & Yoder, 2006). The base rate of the coded behaviors (i.e., proportion of seconds in which each observer recorded each behavior) influences kappa, so that even highly accurate coders can obtain lower values of kappa if the behaviors' base rates are highly unequal. Therefore, coders accuracy was estimated by taking into consideration the base rate of JE and the obtained Kappa (Bruckner & Yoder, 2006).

In the current study, the obtained kappa value was 0.85, regardless of which coder was used as the reference during events alignment. The maximum kappa ranged from 0.99 to 1. Coders' accuracy ranged between 0.95 and 0.99.

2.4.2. Age changes in the amount of time infants were in JE

To determine how the amount of time infants was in JE changed with age, the percentage of time in JE was calculated for each video using Interact (http://www.mangold-international.com). Percentages were calculated separately for: Toys 1: percentage of the first 15 min, when infants used their own toys; and Toys 2: percentage of the second 15 min, when infants used the standard set of toys. To differentiate results depending on the length of JE, percentages were calculated separately for episodes of any duration (following this study's protocol), and those that lasted at least 3 s (following Bakeman & Adamson, 1984). The average and standard deviation of the percentage of time in JE were calculated separately for ages 1 and 2. To determine if the difference in the mean percentages at age 1 versus age 2 were significant, two-tailed *t*-tests with Welch's correction to degrees of freedom were calculated. The Welch's correction, which allows for unequal variance, was used given the unequal number of infants in each age group.

2.4.3. Relationship between visual levels and infants' time in JE

To determine if there was a significant relationship between the infants' visual function and the amount of time they participated in JE, regression analysis was calculated with infants' age, VA, and CS as independent variables, with percentage of time in JE as the dependent variable.

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. For example, normal VA of 20/20 corresponds to logMAR = 0; 20/200 acuity corresponds to logMAR = 1 (a difference of 10 lines on the eye chart between the person's acuity and norm). Adult levels of VA are reached by the end of the first year of life when measured with VEP but not until 3 years of age when measured with PL. Since VA norms vary depending on age, each infant's reduction in VA from the age norm was calculated (in logMAR) for each of the two procedures (VEP and PL). CS has a faster rate of development than VA, reaching near adult levels by one year of age with VEP and PL. CS values were expressed as the lowest detectable contrast in percentages. The contrast levels on the VEP test targets ranged from 80% (nearly black and white) to 1% Michelson contrast. The contrast levels on the PL targets (Mr. Happy test) ranged from 80% to 1.6% (Fisher, Orel-Bixler, & Bailey, 1995).

To determine if there was a significant difference in the VI levels represented at ages 1 and 2, two-tailed *t*-tests with Welch's correction to degrees of freedom were calculated for each of the four visual measurements —VA and CS results based on VEP and PL. To determine if the results from the VEP and PL were correlated, correlations within contrast and acuity measures were computed using Spearman's rho, as these measures did not appear Normal based on QQ-plots (except the preferential looking measure of acuity, which appeared approximately Normal).

All calculations and statistics were computed using R (The R Foundation for Statistical Computing, Vienna, Austria).

3. Results

3.1. Age changes in the percentage of time infants were in JE

All VI infants tested, both the younger and older age groups, participated in JE.

Toys 1 (infants' own toys): When following Bakeman & Adamson's original protocol (1984), only JE events that lasted 3 s or more were considered, a significant difference was found between age 1 ($M = 26.93$, $SD = 15.77$, $N = 9$) and age 2 ($M = 43.04$, $SD = 23.32$, $N = 18$), two-tailed *t*-test with Welch's correction to degrees of freedom $t(22.44) = -2.12$, $p = 0.045$ (Fig. 3).

When, following the current study's coding rule, JE events of any duration were considered, the average percentage of time in JE at age 1 ($M = 27.67$, $SD = 15.87$, $N = 9$) was also significantly different than the average at age 2 ($M = 43.85$, $SD = 22.90$, $N = 18$), two-tailed *t*-test with Welch's correction to degrees of freedom $t(22.07) = -2.14$, $p = 0.043$ (Fig. 4).

Toys 2 (standard set of toys): When only JE events that lasted 3 or more seconds were considered, no significant difference was found between age 1 ($M = 26.41$, $SD = 16.48$, $N = 9$) and age 2 ($M = 35.95$, $SD = 22.70$, $N = 18$), two-tailed *t*-test with Welch's correction to degrees of freedom result $t(21.34) = -1.24$, $p = 0.227$. Likewise, when JE events of any duration were considered, no significant difference was found between age 1 ($M = 27.09$, $SD = 16.78$, $N = 9$) and age 2 ($M = 37.11$, $SD = 22.49$, $N = 18$), two-tailed *t*-test with Welch's correction to degrees of freedom $t(20.89) = -1.30$ $p = 0.208$.

Given the different results obtained with toys 1 and 2, it is important to mention that at age 1 the average percentages of time in JE with toys 1 and toys 2 were similar, whereas at age 2 there was a decrease from toys 1 to toys 2. The change in the percentage of time in JE from toys 1 to toys 2 for age 1 ($M = 0.58$, $SD = 16.60$) and age 2 ($M = 6.74$, $SD = 19.55$) were not significantly different, two-tailed *t*-test with Welch's correction to degrees of freedom $t(18.71) = -0.86$, $p = 0.403$. Since the order of toys were not counterbalanced, it is not possible to determine what drives the difference in results between the two toy sets; factors could include: type of toys, subjects' fatigue, different amounts of caregivers' scaffolding between ages.

Even though no significant difference was found in the percentage of time in JE with toys 2 between ages 1 and 2, collapsing across the ages, there is a positive correlation between the percentage of time between the two sets of toys, Pearson test $r = 0.37$, $p = 0.001$. This suggests that infants who in relation to their age group performance, had a high percentage of time in JE with toys 1,

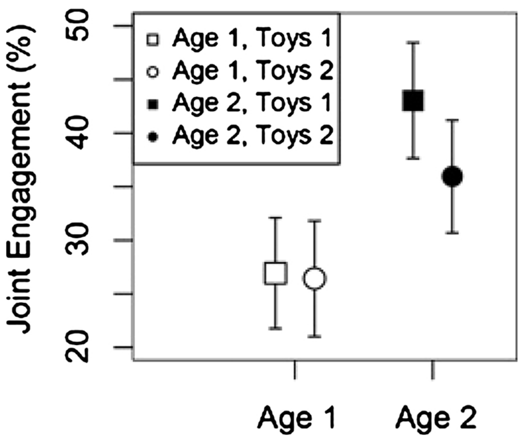

Fig. 3. Joint engagement of 3+ seconds duration (with SEMS) at ages 1 and 2 with toys 1 and toys 2. Toys 1: $t(22.44) = -2.12, p = 0.045$. Toys 2: $t(21.34) = -1.24$, $p = 0.227$.

Fig. 4. Joint engagement of any duration (with SEMS) at ages 1 and 2 with toys 1 and toys 2. Toys 1: $t(22.07) = -2.14$, $p = 0.043$. Toys 2: $t(20.89) = -1.30$ $p = 0.208$.

also had a high percentage with toys 2.

Given that a significant change in average percentage of JE (with toys 1) was found regardless of the exclusion of episodes shorter than 3 s, the following data analysis was done considering JE events of any length.

*3.2. Relationship between visual levels and infants' **time in JE***

3.2.1. Infants' visual levels

The average VA reduction from norm based on the VEP at ages 1 ($M = 0.55$, $SD = 0.31$, $N = 9$) and 2 ($M = 0.42$, $SD = 0.21$, $N = 18$) were not significantly different, two-tailed *t*-test with Welch's correction to degrees of freedom $t(11.84) = 1.14$, $p = 0.27$. Likewise, the average VA reduction from norm based on PL at ages 1 ($M = 0.54$, $SD = 0.67$, $N = 9$) and 2 ($M = 0.44$, $SD = 0.60$, $N = 18$) were not significantly different, two-tailed *t*-test with Welch's correction to degrees of freedom $t(14.09) = 0.36$, $p = 0.72$. The average CS reduction from norm based on the VEP at ages 1 ($M = 5.13$, $SD = 5.41$, $N = 9$) and 2 ($M = 2.43$, $SD = 2.43$, $N = 18$) were not significantly different, two-tailed *t*-test with Welch's correction to degrees of freedom $t(9.66) = 1.42$, $p = 0.186$. Likewise, the average CS from norm based on PL at ages 1 ($M = 29.55$, $SD = 37.94$, $N = 9$) and 2 ($M = 14.23$, $SD = 25.85$, $N = 18$) were not significantly different, two-tailed *t*-test with Welch's correction to degrees of freedom $t(11.84) = 0.78$, $p = 0.297$. These results suggest that the levels of VI of both VA and CS represented at ages 1 and 2 are comparable.

3.3. Models of JE

3.3.1. Participants and age

Some participants were longitudinal, and were therefore represented twice in the data, at both ages 1 and 2. Therefore, we investigated whether a random or fixed intercept for participant would be necessary, and found that neither was necessary. Models with random intercepts for participants were not better than intercept-only models for toys 1 $X^2(1) < 0.01$, $p = 0.999$; nor toys 2 $X^2(1) = 0.99$, $p = 0.319$. Models with fixed intercepts for participants were also not better than intercept-only models for toys 1 $F(19,7) = 0.63$, $p = 0.80$; nor toys 2 $F(19,7) = 2.33$, $p = 0.128$. Thus, intercepts for participants were not included in further models. Age was also not a significant linear predictor of JE, compared to an intercept-only model, for toys 1 $F(1,25) = 1.94$, $p = 0.176$, nor for toys 2, $F(1,25) = 1.37$, $p = 0.254$, and was therefore not included in further models.

3.3.2. Independent variables: acuity and contrast

Collapsing across ages, there is a strong positive correlation in the two acuity measures (VEP and PL), Spearman's rho correlation, $r = 0.80$ (95% CI: 0.61, 0.90), $t(25) = 6.77$, $p < 0.001$. Likewise, collapsing across ages, there is a strong positive correlation in the two contrast measures (VEP and PL), Spearman's rho correlation, $r = 0.74$ (95% CI: 0.47, 0.87), $t(25) = 5.45$, $p < 0.001$). Based on the high correlations between acuity and contrast measures (Spearman's rho correlation values ranging from $r = 0.54$, $p = 0.005$ to $r = 0.82$, $p < 0.001$), it was likely that including all four variables, two for contrast and two for acuity, in a model that predicts joint engagement would produce problems with collinearity (this was confirmed through inspection of the design matrix's eigenvalues, several of which were close to zero). Therefore, forward model selection was used to build a predictive model of joint engagement (%), adding predictors in an order based on relative importance. Relative importance was calculated as R2 partitioned by averaging sequential sums of squares over all orderings of regressors, as in Lindeman, Merenda, & Gold (1980, p. 119 ff), implemented in Grömping (2006; Table 3).

For both sets of toys, the selected model of joint engagement included the single predictor of Contrast PL (Table 4). These models were significantly better than intercept-only models. Adding any of the other predictors to the models did not improve the model fits based on F-tests (all $p > 0.505$).

Table 3
Relative Importance of Acuity and Contrast Measures for Predicting Joint Engagement.

	Toys 1	Toys 2
Acuity VEP	4.24×10^{-2}	2.40×10^{-2}
Acuity PL	4.45×10^{-2}	2.70×10^{-2}
Contrast VEP	4.24×10^{-2}	2.51×10^{-2}
Contrast PL	16.25×10^{-2}	10.30×10^{-2}

To confirm that these results would not have changed based on the order of added predictors, models of joint engagement were constructed using only Acuity PL, which had the second greatest relative importance after Contrast PL, and these models were not significantly better than intercept only models: for toys 1, $F(1,25) = 3.39$, $p = 0.077$; for toys 2, $F(1,25) = 1.81$, $p = 0.190$.

4. Discussion

With the emergence of JE, children start to note how they and others perceive and relate to the same objects/events in a shared physical environment. This allows for other people's engagement with the object to influence the child's relationship with it. As others verbally reference the object and use it in culturally appropriated ways, the child's engagement with it expands beyond exploring the object's physical features and incorporates the object's verbal labels and cultural uses. As social partners call the child's attention to a common focus and respond to the child's calls to attend to a common focus, the child experiences other people as beings with whom he/she can share his/her perceptual experiences. Given this significance, it is regrettable that very limited research has been conducted on JE in VI infants, particularly considering the risks that VI introduces in JE emergence. VI can disturb an infant's contingent interactions with social partners and advances on object knowledge, which are necessary for JE. VI infants may show slower patterns of JE development, which are deemed "atypical" and considered early signs of autism (Hobson, 2005). However, atypicalities in this population of infants cannot be determined by comparing them to sighted peers' norms. JE development in infants with degraded or absent vision will logically differ from sighted peers in its mechanisms and timelines. Failing to recognize this puts VI children at risk of being wrongly labeled as autistic. It is thus necessary to characterize JE development in this population. Furthermore, this research is relevant to theories of JE in sighted children. Current accounts heavily assume infants' normal visual abilities. As a consequence, little is known about the development of JE when vision is atypical, and theories assume, perhaps incorrectly, that normal visual experiences are required for the optimal development of JE.

The current study found that all of the VI infants tested participated in JE which suggests that VI infants can show joint engagement around age 12-month. We use "can" when stating this finding because all of the sample's infants were receiving developmental counseling. The way VI impacts children's development does not solely depend on the sensory limitation itself, but also on the degree caregivers and society accommodate to these children's needs and strengths. It is possible that studies with VI infants who are not receiving developmental counseling could find a later age onset than what we report. Given our findings however, we argue that a slower pattern of JE development is not an unavoidable consequence of VI.

Likewise, it is important to note that our results cannot be extrapolated to the population of totally blind children. As it is the case with the majority population of VI children, most participants in our sample had residual vision. Furthermore, our focus was on these children's participation in JE with their main caregivers, irrespective of the senses, visual or non-visual. Thus, our coding system did not specify the senses involved in the observed JE and our findings do not report on how VI children enter into joint engagement through non-visual means only.

Even infants with the most severe VI levels represented in our sample participated in JE. However, given our focus in total joint engagement, our results do not disprove previous studies that found greater difficulties in CJE in children with severe VI. Given the lower attention coordination requirements for participating in supported versus coordinated JE, VI may introduce lower risks to the emergence of SJE, compared to CJE. SJE requires the social partner to monitor and direct the child's attention to a shared focus. Even if caregivers have difficulties identifying the focus of their VI child's attention, they can direct the child's attention to an object. For

Table 4
Models of Joint Engagement.

Predictor	Toys 1			Toys 2		
	Est. (SE)	t-value	p-value	Est. (SE)	t-value	p-value
Intercept	45.54 (4.40)	10.34	< 0.001 ***	39.04 (4.50)	8.68	< 0.001 ***
Contrast PL	−0.37 (0.12)	−2.97	0.006 **	−0.27 (0.13)	−2.16	0.040 *
	$F(1,25) = 8.81$, $p = 0.006$ $R^2 = 0.26$, Adj. $R^2 = 0.23$			$F(1,25) = 4.67$, $p = 0.040$ $R^2 = 0.16$, Adj. $R^2 = 0.12$		

* $p < 0.05$.
** $p < 0.01$.
*** $p < 0.001$.

CJE however, redirecting the child's attention to a shared focus is not sufficient. CJE requires that the caregiver and child both acknowledge each other's involvement in the engagement of the shared focus. Since we did not differentiate between SJE and CJE, we think it is important to stress that our results do not address the more circumscribed question of CJE development in VI infants.

Our results showed that infants in the older age group participated in joint engagement in a significantly higher percentage of time than infants in the younger age group. This suggests that a characteristic of JE development in VI infants who receive developmental services is a significant increase in the percentage of time they are in JE with their main caregivers, as they age, with an important change occurring roughly between the end of the first year and the middle of the second year of life. It is interesting to note that this pattern of development has been documented in sighted children (Adamson et al., 2014).

It is relevant to consider the free play context in which we measured JE. Data collected in this way tell us about the amount of JE these infants and their caregivers participate in when left to their own devices. This had the benefit of reducing the risk, introduced by prompts in structured play contexts, of over-estimating the amount of JE in naturalistic interactions. By the same token, it could have underestimated the infants' JE. In all children, but particularly in the case of VI infants who manifest attention in idiosyncratic ways, participation in JE can vary depending on the social partner's ability to interpret and influence the infants' attentional states. Our findings cannot be extrapolated to interactions with unfamiliar people such as developmental evaluators meeting these infants for the first time.

We previously warned against assessing the normalcy of VI infants' development by comparing it that of sighted peers. Nonetheless, comparing this study's findings to those previously reported in sighted infants can inform theories of JE in sighted children. Current accounts consider infants' visual perceptions of social partners relating to objects as the source of information that most efficiently contributes to JE. Seeing others' facial/body changes to objects would allow children to perceive others' relatedness to a shared world in the most salient and least ambiguous way. We can investigate this idea by comparing JE development in VI and sighted children. Evidence of better JE developmental outcomes (e.g., earlier age onset, higher percentages of JE) in sighted children would support the facilitative role of vision. Results that rather show similarities in outcomes between the two populations would suggest that infants' abnormal vision does not lead to less optimal development of JE.

Bakeman and Adamson's results (1984) are the most appropriate to compare to ours since they also used free play, and our study largely followed their coding protocol—although they had 10-min rather than 30-min play sessions as in our study. Since we adapted their coding protocol however, comparisons are not straightforward. As explained earlier, some of the episodes we coded as JE could be coded as "onlooking", a type of non-JE, according to the original protocol. Thus, if we compare our results to Bakeman and Adamson's percentages in total JE (summing SJE and CJE, without including onlooking), we may be underestimating the differences between the samples. At the same time, not all of the episodes that could be coded as onlooking in the original protocol would be coded as JE according to our adapted protocol (e.g., if the infant was merely looking at the caregiver's action with the object, without the caregiver having called the infant's attention to the object, this was not coded as JE). Thus, if we compare our results to the sum of Bakeman and Adamson's percentages in total JE and in onlooking, we may be overestimating the differences between the two samples. Thus, below we provide both types of comparisons. We only considered JE episodes that lasted at least 3 s since that was Bakeman and Adamson's criterion.

In Bakeman and Adamson's study, 12-month-old sighted infants were in JE for an average of 22.9% of the time. If onlooking is included, the JE percentage increases to 35.8. In our study, VI infants with mean fractional age of 1.08 were in JE a mean time of 26.93% with the infants' own toys, and 26.41% with the standard toys. Thus, our sample showed similar (yet higher) percentages of time in JE around the end of the first year of life, but only if onlooking is not included in the comparison. If onlooking is included, our study's VI infants were in JE a comparatively lower percentage of time. In Bakeman and Adamson's, the mean JE percentage for 18-month olds was 48.1%. If onlooking is included, the percentage increases to 55.6%. In our study, VI infants with a mean fractional age of 1.62 were in JE 43.04% of the time with their own toys, and 35.95% with the standard toys. At this older age, comparisons differ by the type of toy. When using their own toys, VI infants showed a similar (but this time lower) percentage in JE. As at 12-months, this was the case only if onlooking was excluded; when onlooking is included, VI infants showed a lower JE percentage. When using the standard toys, VI infants at the older age were in JE a lower percentage of time whether onlooking is included or not.

Given CS's relationship to environmental navigation and perception of facial expressions, we suspected that a child who is better able to respond to brightness differences will likely have greater opportunities to perceive objects and people's attentional cues, and thus access more information conducive to JE. A child who detects more useful visual information for navigation (i.e., differences in surface's depth) will be more likely to venture into the environment and encounter and re-encounter objects in diverse space locations. This contributes to the child's understanding that objects exist independent of their location in space, and provides for more opportunities to encounter objects that can be the focus of JE. A child who can better discriminate facial expressions will experience social partners' faces as a more salient stimulus, and detect more of the partner's facial responses to objects and infant's behavior. These abilities are conducive to the child engaging in contingent social interactions and noticing how social partner's behaviors relate to a shared external world.

Indeed, infants' CS levels, measured with PL (but not VEP), predicted the percentage of time VI infants in our sample spend in JE. We propose the following explanation to CS predicting JE when measured with PL alone. Since the VEP gives general information about the occipital response to visual stimuli and the PL reports behavioral use of vision, this finding suggests that the critical factor for JE in VI infants is the degree to which they utilize input on differences in brightness in their behavioral responses. It should be noted that the PL test used in the current study was limited in that it did not include steps between 25% and 80%. This limitation is shared by other commercially available PL tests of CS in infants. In future research, investigators should include CS measurements, and test contrast values between 25% and 80%.

In discussing this study's results, we have mentioned several study limitations. Because all infants in our sample were receiving developmental counseling and had residual vision, our findings cannot be extrapolated to VI infants that lack developmental counseling nor to the population of infants that are totally blind. Our coding protocol did not differentiate between SJE and CJE and thus our results do not disprove previous research that has reported VI children's difficulties in CJE. To these limitations, we would like to add that, even though our sample size of 20 VI infants without additional disabilities is relevant given how challenging it is to collect data from this low incidence population, research with larger samples is needed.

In summary, this investigation shows that VI infants, without additional disabilities, whose families receive developmental services, can participate in JE with their main caregivers toward the end of the first year of life. JE in VI infants continues to develop after its emergence, increasing significantly between roughly 12 and 19 months of age. VI infants' behavioral use of differences in brightness input predicted JE. These findings contribute to the characterization of the typical development of JE in VI infants and how it relates to these infants' visual levels.

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