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Language and affective facial expression in children with perinatal stroke

Philip T. Lai [a,b,*,1], Judy S. Reilly [a,c,2]

[a] San Diego State University, United States
[b] University of California San Diego, United States
[c] Université de Poitiers-CNRS, France

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ABSTRACT

Children with perinatal stroke (PS) provide a unique opportunity to understand developing brain-behavior relations. Previous research has noted distinctive differences in behavioral sequelae between children with PS and adults with acquired stroke: children fare better, presumably due to the plasticity of the developing brain for adaptive reorganization. Whereas we are beginning to understand language development, we know little about another communicative domain, emotional expression. The current study investigates the use and integration of language and facial expression during an interview. As anticipated, the language performance of the five and six year old PS group is comparable to their typically developing (TD) peers, however, their affective profiles are distinctive: those with right hemisphere injury are less expressive with respect to affective language and affective facial expression than either those with left hemisphere injury or TD group. The two distinctive profiles for language and emotional expression in these children suggest gradients of neuroplasticity in the developing brain.

1. Introduction

As early as the mid-eighteenth century it had been proposed that early brain injury would lead to more favorable outcomes than similar injury occurring later in life. Empirical studies later confirmed this hypothesis (Basser, 1962; Bates et al., 2001; Lenneberg, 1967; Reilly, Levine, Nass, & Stiles, 2008; Stiles, Reilly, Levine, Trauner, & Nass, 2012). Such positive results are generally attributed to neuroplasticity, the developing brain's ability to flexibly adapt and reorganize (Cao, Vikingstad, Huttenlocher, Towle, & Levin, 1994; Chu, Huttenlocher, Levin, & Towle, 2000; Kirton & Deveber, 2006; Stiles et al., 2012). Evidence of brain plasticity stems from animal studies in a variety of areas: the motor cortex (Kennard, 1936, 1942), visual systems (Hubel & Wiesel, 1967; Hubel, Wiesel, & LeVay, 1977; Wiesel, 1982; Wiesel & Hubel, 1963, 1965), enhanced performance in enriched environments (Greenough & Chang, 1989; Hebb, 1947; Kempermann & Gage, 1998; Kempermann, Kuhn, & Gage, 1997; Rosenzweig & Bennett, 1972), the successful rewiring of cortical projections (Angelucci, Clasca, Bricolo, Cramer, & Sur, 1997; Neville, Schmidt, & Kutas, 1983; Sur, Garraghty, & Roe, 1988), and finally the development of transplanted fetal tissue (O'Leary and Stanfield, 1989).

While we have learned much about neuroplasticity from animal models, children with perinatal stroke (PS) offer an opportunity to better understand the nature and extent of neuroplasticity in humans. Looking across different cognitive domains in children with PS has yielded differing results (see Stiles et al., 2012 for a comprehensive review). For example, group studies of language in children with PS have shown initial delays in the onset of language regardless of lesion site (Bates et al., 2001; Rowe, Levine, Fisher, & Goldin-Meadow, 2009; Thal et al., 1991; Vicari et al., 2000). However, by middle childhood, spontaneous language is in the low-normal range (Bates et al., 2001; Reilly, Bates, & Marchman, 1998; Reilly, Losh, Bellugi, & Wulfeck, 2004). As such, the language profile of children with early stroke does not mirror that of adults with homologous lesions. Emotion is another communicative system. In contrast to the profile for early language, the few studies on emotion with infants and toddlers with PS (e.g., Nass & Koch, 1987; Reilly, Stiles, Larsen, & Trauner, 1995) have shown that the site of injury is associated with differential profiles of expressiveness and emotionality. Moreover, the infant profile is similar to that of adults with late onset strokes (Borod, Koff, Lorch, & Nicholas, 1985; Borod et al., 1998). To better understand the nature of neuroplasticity in the developing brain, the present study investigates language and emotional expression in

* Corresponding author at: San Diego State University, 6330 Alvarado Court, #208, San Diego, CA 92120, United States.

E-mail addresses: ptlai@ucsd.edu (P.T. Lai), reilly1@mail.sdsu.edu (J.S. Reilly).

[1] SDSU/UCSD Joint Doctoral Program in Language and Communicative Disorders, San Diego, CA, United States.

[2] Department of Psychology, San Diego State University, San Diego, CA, United States.

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young school age children with PS. Will their profile of affective expressivity follow that of language, such that by school age, the children with PS perform like their typically developing (TD) counterparts, regardless of lesion site? Or will the emotion profile continue to be characterized by site-specific deficits mirroring that of adults with acquired strokes? To address these questions, we investigate the production of emotional expression in both language and facial expression, as well as their integration during a semi-structured naturalistic biographical interview. Together these results will enhance our understanding of both the nature and extent of neuroplasticity in the developing brain.

1.1. Language development in children with perinatal stroke

Jules Cotard, a colleague of Paul Broca, suggested that unlike an adult with a comparable lesion, a child with an early left hemisphere stroke would not be aphasic (Cotard, 1868; Levin & Grafman, 2000). The reports of Lenneberg (1967) and Basser (1962) confirmed this observation leading to Lenneberg's hypothesis of equipotentiality, that is, that either hemisphere of the brain could assume language functions. More recently, prospective studies of children with PS have provided an opportunity to understand the dynamics of early brain injury and their behavioral and cognitive sequelae (e.g. Ballantyne, Spilkin, Hesselink, & Trauner, 2008; Chilosi et al., 2005, 2008; Feldman, 2005; Reilly et al., 1998, 2004; Rowe et al., 2009; Stiles, Stern, Trauner, & Nass, 1996; Stiles et al., 2012; Yeatman & Feldman, 2013). Within the context of language development, studies have found distinctively different outcomes for children and adults with analogous lesions. In the adult language profile, the side, site, and the size of the lesion is associated with particular types of deficit. Studies from a broad range of languages have shown that adults with left hemisphere strokes are often aphasic (Broca, 1861; Goodglass, 1993; Goodglass & Hunter, 1970; Wernicke, 1874), whereas those with right hemisphere injury tend to be garrulous, and show deficits in discourse cohesion and in processing non-literal language (Abusamra, Côté, Joanette, & Ferreres, 2009; Joanette & Goulet, 1990, 1994; Joanette, Goulet, Ska, & Nespoulous, 1986; Lundgren, Brownell, & Keith, 2006). Imaging studies of healthy adults and neurological patients broadly confirm that for adults, a network subsuming left perisylvian regions is implicated in processing core aspects of language (Bookheimer, 2002; Brauer, Anwander, & Friederici, 2011; Price, 2010; Tyler & Marslen-Wilson, 2008; Tyler, Stamatakis, Post, Randall, & Marslen-Wilson, 2005). Importantly, young children with PS do not show the same pattern of linguistic deficits as adults with comparable lesions. Specifically, young children with either left or right hemisphere lesions present with delays in the major milestones of language acquisition: babbling, gestures, and both comprehension and production of first words (Bates et al., 1997; Marchman, Miller, & Bates, 1991; Reilly et al., 2008; Sauer, Levine, & Goldin-Meadow, 2010; Stiles, Bates, Thal, Trauner, & Reilly, 1998; Thal et al., 1991; Vicari et al., 2000). Within this profile of overall delay, several studies have found increased production deficits for those toddlers and preschoolers with left hemisphere injury (Bates et al., 1997; Chilosi, Cipriani, Bertuccelli, Pfanner, & Cioni, 2001; Vicari et al., 2000) whereas comprehension deficits were reported in toddlers with right posterior injury (Bates et al., 1997). Interestingly, comprehension deficits were not found in children with injuries to left temporal areas, as the adult profile would have predicted. Rather, those with left temporal injury showed delays in word production (Bates et al., 1997). Thus, during the early stages of language development, the children's profiles do not map onto those of adults with homologous injuries. Moreover, the pattern of deficits strongly suggests that acquiring language, as opposed to maintaining a mature system, requires both the right and left cerebral hemispheres (Reilly et al., 2008). Imaging studies have lent support to this view: language processing in TD children is bilaterally mediated and left hemisphere specialization for language is a prolonged developmental process (Brown et al., 2005; Holland et al., 2001).

Remarkably, by the time children with PS enter school, their spontaneous language performance is broadly comparable to their TD peers. Bates et al. (2001) used speech samples from biographical interviews to examine spontaneous language production in brain-injured children and adults with comparable lesions. The adult stroke patients showed classic hemispheric differences with regards to language; none of these characteristics were found in children with early brain injury (ages 5–8). Overall the children with PS were in the normal range, that is, within one standard deviation of the normal mean for their ages, on all comparisons including frequency of morphological errors and use of complex syntax (Bates et al., 2001). In addition, performance of those children with left hemisphere injury (LHI) was comparable to that of children with right hemisphere injury (RHI). In sum, although there is an initial delay in reaching the early milestones of language acquisition for those with either LHI or RHI, children with PS eventually develop functional language and perform within the low-normal range in spontaneous speech by school age (Bates et al., 2001; Reilly et al., 1998, see Stiles et al., 2012 for a review).

This pattern of delay and development has also been demonstrated in oral picture story narratives. For example, Reilly and colleagues found that children with RHI or LHI were initially delayed on grammatical and narrative measures (Reilly et al., 1998, 2004). In the youngest group, ages 4–6, children with PS told shorter stories, made more morphological errors, produced fewer complex sentences, and included fewer story components in their narratives than did their TD peers. But by age 10, the PS group performed in the low-to-normal range on all morphosyntactic as well as narrative discourse measures, and there were no significant hemispheric differences.

These differential results in conversation and narratives point to the importance of discourse context in the linguistic performance of children with perinatal stroke. Some of the same children (ages 5–8) participated in both the conversational (Bates et al., 2001) and narrative studies (Reilly et al., 1998, 2004). In the conversation, their performance was in the normal range; in the picture story narrative, a cognitively more challenging task, the linguistic performance of these younger children with PS (both RHI and LHI) fell below their TD peers. In a complementary narrative study with 5–7 year olds with PS that used story stems, Demir, Levine, and Goldin-Meadow (2009) also found more impoverished narratives from the PS group. To investigate both later language development and how context influences performance, we asked children with PS (ages 7–16) to recount a personal narrative in response to the prompt, "Tell me about a time someone made you sad or mad," (Reilly, Wasserman, & Appelbaum, 2012). In this case, not only is the child/adolescent asked to create her own story (as opposed to the pictures presenting the story), but the additional emotional component further increases the cognitive challenge. In this particular context we found that the language performance of the children with LHI trailed that of the TD group in both morphosyntactic and discourse measures while the performance of the group with RHI was broadly comparable to the TD group.

In summary, after initial delay in the onset of language, children with PS tend to follow a steady developmental progression, acquiring language milestones in a similar sequence to their TD peers and performing within the low-to-normal range in spontaneous language production by middle childhood. Such profiles demonstrate the role of both hemispheres in acquiring language and the