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EARLY LANGUAGE DEVELOPMENT IN EXTREMELY PRETERM INFANTS:
RELATIONSHIPS BETWEEN INFANT COMMUNICATIVE AND MOTOR SKILLS
AND ROLE OF MATERNAL RESPONSES

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ABSTRACT

Despite the importance to identify infants at risk for later language concerns, little research focused on early communicative behaviors in extremely-low-gestational-age infants (ELGA, GA < 28 weeks). In particular, none investigated the multimodal communication in these infants, also considering possible connections to motor skills. Furthermore, rarely the communicative development of these infants has been studied as a result of the interplay among individual and environmental components. Thus, guided by the theoretical framework of the Dynamic Systems Theory, which views development as a result of the interaction between multiple subsystems within the infant and the context, two studies were designed.

In Study 1, spontaneous communicative behaviors (gestures, vocal utterances, and coordinations) were evaluated during mother-infant interactions in 20 ELGA infants and 20 full-term (FT) infants at 12 months. Less advanced communicative and motor abilities emerged in ELGA infants relative to FT infants. Giving and representational gestures were produced at a lower rate by ELGA infants, and pointing gestures and words were produced by a lower percentage of ELGA infants. Positive associations between gestural and fine motor skills were found in the ELGA group.

In Study 2, maternal responses provided to the infants' communicative behaviors were coded with regard to contingency and relevancy at 12 months. The mothers of the ELGA infants did not

appear at risk for providing less prompt and meaningful responses relative to the mothers of FT infants, and their relevant responses were strictly related to their infants' communicative abilities at 12 months. Overall, the repeated labeling responses had a predictive effect on the expressive language at 24 months.

We discuss the importance to combine spontaneous communicative behaviors and motor skills in the clinical assessment and early intervention with ELGA infants. We also emphasize the usefulness of the maternal repeated labeling for supporting language development of these infants.

Keywords: Extremely preterm birth, communicative behaviors, motor skills, maternal responses, language development

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PREFACE

I still remember the words of the supervisor of my master thesis, Prof. Pierluigi Zoccolotti: “I’m pleased you want to do the Ph.D.; among all my colleagues, in fact, I prefer who are both speech therapists and researchers”. Thus, I started the Ph.D. with enthusiasm putting together the research I did and my clinical expertise. I am pleased to extend my thanks to all people who have instilled in me to continue working in this direction.

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1. INTRODUCTION

1.1 Focusing on extremely preterm birth

According to the World Health Organization (WHO, 1970; Villar, Betrán, Gulmezoglu, & Say, 2003), preterm birth is defined as being born before 37 weeks gestational age or before 259 days. This definition includes a widely heterogeneous population, because of large variations depending on the degree of neonatal immaturity, medical complications, and environmental agents.

Regarding the degree of neonatal immaturity, it is determined by birth weight (BW) and gestational age at birth (GA). With regard to BW, preterm infants are distinguished in three main categories: low birth weight (LBW, <2,000 g), very low birth weight (VLBW, <1,500 g), and extremely low birth weight (ELBW, <1,000 g) preterm infants (Goldenberg, Culhane, Iams, & Romero, 2008). In the last 20 years, more attention has been given to GA at birth that appears the main index of the level of physical and neurological maturation and it is now recognized as a reference standard related to the outcome and prognosis of preterm infants (Sansavini, Guarini, & Caselli, 2011). Thus, low-gestational-age preterm infants (LGA, 80%, that is 60% late preterm, born between 34 and 36 weeks GA and 20% mild preterm, born between 32 and 33 weeks), very-low-gestational-age preterm infants (VLGA, i.e. 15%, between 28 and 31 weeks GA) and extremely-low-gestational-age preterm infants (ELGA, i.e. 5%, <28 weeks GA) have been distinguished (Goldenberg et al., 2008). It is clear that preterm infants born before 28 weeks of GA are more

vulnerable than preterm infants born at 31-32 weeks of GA; just consider the fact that most babies born at less than 28 weeks GA need neonatal intensive care services to survive (Blencowe, Cousens, Chou, Oestergaard, Say, et al., 2013). Thus, preterm birth can result in a range of long-term neurodevelopmental effects, such as moderate/severe cognitive and language delays, with the frequency and severity of adverse outcomes rising with decreasing GA and presence of cerebral or neurological damages (Blencowe et al., 2013). Given that the main studies of neuropsychological development in the preterm population have recruited preterm infants with a wide range of GA or have included healthy as well as neurologically damaged infants, not always they are easily comparable and show conflicting results (Marlow, Wolke, Bracewell, Samara, & EPICure Study Group, 2005). To better understand the effects of preterm birth on development, a first challenge for the researcher is therefore to collect homogeneous samples of preterm infants.

To increase our understanding about the effects of preterm birth on development, it is also important to consider environmental factors that play a role in neuropsychological development of preterm infants. For example, as highlighted by Blencowe and colleagues (2013), frequency and severity of adverse outcomes rise also with decreasing quality of care; thus, the characteristics of the neonatal units in which the preterm infants receive the first care should be considered. Also the communicative feedbacks that preterm infants receive by the adults around them may influence the development of these infants; the cultural and linguistic background and the interventions offered to the preterm infants, should be taken into account as well (Sansavini, Guarini, & Caselli, 2011). Therefore, complex interactions between bio-medical and socio-environmental factors may concur in determining developmental risk.

With the purpose to reduce as much as possible this heterogeneity of factors involved, the present study focused on a homogeneous group of ELGA infants: all infants were born less than 28 weeks of GA, were healthy (i.e. without cerebral and neurological damages), and were born at the Neonatal Intensive Care Unit (NICU) of Bologna University from monolingual Italian parents.

ELGA infants are scrawny and their body at birth is only a little larger than the size of an hand (i.e. birth weight of 500-1500 grams and birth length of 35-40 cm; Douglas & Pearson, 1971; Fenton, 2003); they are cared for in a special incubator, breath with the aid of a respirator, fed through a stomach tube, and receive medication through an intravenous needle during the first postnatal weeks (Berk, 2010).

ELGA infants are at highest risk for medical complications, such as periventricular leukomalacia, respiratory distress syndrome, bronchopulmonary dysplasia, intra-ventricular hemorrhage, retinopathy of prematurity, and hyperbilirubinemia (Aylward, 2009). ELGA infants are also at highest risk for cerebral damages (Asl, Duffy, McAnulty, Rivkin, Vajapeyam, et al., 2004) and for developmental delays (Sansavini, Guarini, & Caselli, 2011).

In addition to all these bio-medical risk factors, socio-environmental factors concur to determine the developmental risk of ELGA infants (Aylward, 2009). The extremely preterm birth is a stressor event for the parents (Trombini, Surcinelli, Piccioni, Alessandroni, & Faldella, 2008); the mothers of ELGA infants, in particular, often develop psychological distress and are worried about the development of their infants, with negative effects on the early mother-infant interaction (Bozzette, 2007; Coppola, Cassibba, & Costantini, 2007; Trombini et al., 2008). Other environmental factors, such as the education level and the socio-economic status of the parents appear to affect the developmental outcomes of the ELGA infants with more evident effects from second-third year of life (Aylward, 2009; Sansavini, Guarini, Justice, Savini, Broccoli, et al., 2010). The ELGA infants development can thus be understood as originating from the interplay of all these multiple biological and environmental constraints (Sansavini, Guarini, & Caselli, 2011).

It is evident that the extremely preterm birth is a traumatic event in the life of these infants and their families. According to a recent publication of the World Health Organization (Villar et al., 2003), about 13 million babies are born preterm annually (Beck, Wojdyla, Say, Betran, Merialdi, et al., 2010), and medical progress has improved the survival rate of ELGA preterm newborns to 70-

80% (Saigal and Doyle, 2008). Therefore, nowadays, focusing on ELGA infants and their mothers assumes scientific and clinical relevancy.

1.2 Purposes of the present research

The present work was guided by two research questions regarding early communication development in ELGA infants and two research questions concerning the role of maternal communicative input on infant's communicative development. Specifically, the following research questions were posed: 1) Do ELGA infants show less advanced multimodal communicative abilities, compared with FT infants, at 12 months of age? 2) Are the communicative and motor domains related in the ELGA group? 3) Do the mothers of ELGA infants differ from the mothers of FT infants in the way in which they respond to their infants' communicative behaviors? 4) Do maternal responses influence the communicative-linguistic development of ELGA infants?

With the objective to thrash out these issues, this research is comprised of two studies designed to examine: 1) the early communicative behaviors exhibited by ELGA infants, compared with those of full-term (FT) infants, and their relationship to early motor skills; 2) maternal responses to ELGA and FT infant's communicative behaviors and their influences on language development of these infants.

The conceptual framework employed in this study was based on the Dynamic Systems Theory (DST; Thelen & Smith, 1994; see par. 3.1). The DST view of communicative development involves a collective system with dynamic interplay among multiple components within the infant and the environment, from which the complex phenomena development (e.g., language) depends. Thus, to better understand language development of ELGA infants, there is a need to examine the multimodal communication abilities that these infants exhibit at early age relative to FT infants, and their relationships both with other domains, such as the motor domain, and with the environmental

factors, such as the maternal responses.

As I will describe in the next sections (see par. 3.2), the literature concerning early communicative behaviors in preterm infants is partially conflicting, probably due to different methodological choices (i.e. sample criteria selection, such as inclusion of preterm infants with a wide range of gestational age and with cerebral damages, and types of measures). A strength of this study is the homogeneity of the sample: healthy ELGA infants were examined, i.e. infants born less than 28 gestational weeks and without cerebral or neurological damages. Among preterm infants, the ELGA infants are those at highest risk for communicative-linguistic delays and impairments (Sansavini, Guarini, et al., 2010). Despite the relevance of identifying early predictors of language delay in at risk populations, for suggesting potential targets for intervention, few studies have been specifically conducted on the early communicative behaviors of ELGA infants. The vast majority of the studies concerned with early communicative development in preterm infants were focused on VLGA infants and on individual communicative behaviors. Moreover, although some authors suggest a strict link between infant language and motor abilities (Iverson, 2010) and some evidence of this relationship was found in typically and atypically developing infants (Iverson, 2010; Leonard & Hill, 2014; see par. 3.3), and despite that ELGA infants show less advanced early motor skills (Sansavini, Pentimonti, Justice, Guarini, Savini, et al., 2014), the relationship between early motor and communicative abilities has not yet been investigated in the ELGA population. Study 1 investigates, for the first time, early multimodal communication abilities (gestural, vocal, and their coordination) of ELGA infants and the relationships between gestural behaviors and motor skills, comparing them to those of FT infants. Most of the studies focused on preterm infants' early communicative development have used indirect tools, such as parental report measures. Thus, another strength of Study 1 was the use of two direct tools (i.e., microanalytic coding of spontaneous communicative behaviors during mother-infant interaction and a standardized test for evaluating infant's abilities), in addition to a parental report measure, to assess communicative

development of the two groups of infants at 12 months.

Consistent with the DST, the examination of environmental factors that could affect the communicative-linguistic development of ELGA infants, such as the maternal feedback to infant's communicative behaviors, may improve our understanding of the mechanisms underlying language acquisition in this clinical population. From a DST perspective, contextual factors interacting with the individual are the critical aspects of performance and development (Smith & Thelen, 2003). Infant's and parent's behaviors are mutually influential and this dynamic interaction affects infant's development in significant ways. From the literature on mothers of preterm infants (see par. 3.4), most studies have focused on maternal communicative strategies without considering the infants' role in shaping their own environment input. The goal of Study 2 was therefore to investigate if the less advanced communicative abilities of the ELGA infants may influence the way in which their mothers respond to their infants at 12 months of age. Specifically, for their importance on language development (Baumwell, Tamis-LeMonda, & Bornstein, 1997; Girolametto, Weitzman, Wiigs, & Pearce, 1999; Tamis-LeMonda, Bornstein, & Baumwell, 2001), we focused on contingency (i.e. the capacity to provide prompt responses to infant's communicative behaviors) and relevancy (i.e. the capacity to provide informative and pertinent responses to infant's communicative behaviors) of the maternal responses. To date, none has considered these types of responses in mothers of ELGA infants, relative to mothers of FT infants. The novel contribution of study 2 was to verify whether maternal relevant responses, particularly those including labeling in response to infant's communicative behaviors, were related to infant's early communicative behaviors at 12 months. Moreover, the second important contribution of this study was to investigate whether maternal labeling responses contribute to predict ELGA infant's language skills assessed at 24 months. The study of the effect of maternal labeling responses on language development has received little attention in studies on preterm infants. We believe that studying these maternal factors can contribute to further understand the language outcomes of this clinical population.

Before proceeding to illustrate the present study, I will provide a brief overview of the current methods for evaluating communicative abilities in infants and maternal communicative responses.

2. METHODS

2.1 Methods for studying infant and maternal communicative behaviors in full-term (FT) and preterm populations

Studying early infant and maternal communicative behaviors according to scientifically accepted procedures involves many steps, such as the choice of measures for examining the behaviors of interest. This choice poses a challenge for researchers. Given the low cooperation that young children in general show during their assessment, it does not appear easy to understand which is the best accurate and informative measure for gathering information at early age. Common methods of detecting information about communicative skills in young children with and without developmental delay include parental report measures, structured assessment tools, and the observation of the spontaneous mother-infant interaction in laboratory semi-structured setting or in naturalistic environment. With regard to maternal communicative strategies, the observation of the spontaneous mother-infant interaction is the most widely employed measure.

2.1.1 Parental report measures

A first approach to measuring infant's early communicative abilities is to use information reported by familiar adults gathered in questionnaire or interview format. Of the currently available assessment tools, the Words and Gestures form of the MacArthur-Bates Communicative

Development Inventory (CDI-WG, Fenson, Marchman, Thal, Dale, Reznick, & Bates, 2007) documents the largest number of early emerging communication skills. The Gestures and Words form of the *Primo vocabolario del bambino* questionnaire (PVB-GW; Caselli & Casadio, 1995) is the Italian version of the CDI-WG, designed for infants from 8 to 17 months. The PVB-GW consists of yes/no questions that investigate infant's receptive/expressive vocabulary (e.g., animals, vehicles, toys, clothings, body parts), gestures (e.g., pointing, giving) and actions (e.g., games and routines, pretend play). Parents are asked to check whether the infant has displayed each behavior. The questionnaire is a reliable and valid parental report measure of communication and action development which has been used extensively in research with typically developing infants (TD) and preterm infants (Ortiz-Mantilla, Choudhury, Leevers, & Benasich, 2008; Sansavini, Guarini, Savini, Broccoli, Justice, et al., 2011). Others types of parental questionnaires have been used to detect early communicative abilities in preterm infants (e.g., see Gonzalez-Gomez & Nazzi, 2012; Torola, Lehtihalmes, Heikkinen, Olsen, & Yliherva, 2012). Thus, to date, the parental questionnaires are the most widely employed measures in studies concerned with early communicative development in preterm infant.

Relative to others types of measurement, parental report measures have some strengths: they capitalize on the knowledge of a familiar person who interacts with the infant on a daily basis; they use the same set of questions in the same way with each participant; filling out by the parents and the scoring by the researcher are simple and fast. Nevertheless, parental report measures do not yield the same depth of information as a direct observation can do, and parent's responses might be subject to inaccurate reporting.

2.1.2 Observation and coding of spontaneous behaviors

A second approach to measuring infant communicative abilities is to observe the spontaneous mother-infant interaction. Frequency, quality, and complexity of infant's communicative behaviors

(e.g., spontaneous gestures and vocal productions) can be sampled in interactive play contexts. The observation of the spontaneous mother-infant interaction can be carry out in laboratory semi-structured setting or in natural contexts (e.g., the home observation). Both in the laboratory semi-structured setting and in the naturalistic home observation, infants and mothers sit together on a mat and play with toys (e.g., car, ball, animal toys) and picture books, as they normally would. Despite many of the spontaneous infant's behaviors are maximally likely to occur when infants are in familiar surroundings with respect to laboratory (Lewedag, Oller, & Lynch, 1994; Iverson, Capirci, & Caselli, 1994), the familiar setting created in the laboratory can promote the exhibition of many spontaneous behaviors. The procedure used to collect systematic observations vary, depending on the research problem posed. Typically, between age 6 months and 4 years, researchers videotape mother-infant 15-minute play session (Berk, 2010) and then they execute an off-line coding of the behaviors of interest using coding schemes or check-list.

The primary advantage of observing spontaneous behaviors is that it reflects participants' everyday lives, and thus provides an ecologically valid measure able to capture a wider repertoire of infant behaviors relative to parental report measures. Observation of spontaneous behaviors is a reliable method that has been widely and successfully employed in numerous studies of early communicative development, both in TD (e.g., Iverson, Capirci, & Caselli, 1994) and atypically developing infants (e.g., Winder, Wozniak, Parladé, & Iverson, 2013). Observation of spontaneous behaviors has also been employed to study the early vocal and gestural productions in VLGA infants (D'Odorico, Majorano, Fasolo, Salerni, & Suttora, 2011; Suttora & Salerni, 2012) and in ELBW infants (Torola et al., 2012), but never in ELGA infants.

Observation of the spontaneous mother-infant interaction has also largely been employed in studies concerning with maternal communicative strategies (e.g., maternal responsiveness), both in TD (Baumwell et al., 1997; Girolametto et al., 1999; Goldin-Meadow, Goodrich, Sauer, & Iverson, 2007; Gros-Louis, West, & King, 2014; Rollins, 2003; Tamis-LeMonda et al., 2001) and atypically

populations (Leezenbaum, Campbell, Butler, & Iverson, 2014).

A limitation of this method is that the coding of the behaviors of interest may take long time, and thus it is not easily practicable in clinical assessment. However, this procedure allows detailed descriptions of frequency and quality of infant and maternal communicative behaviors. By contrast, few studies have employed this type of methodology to investigate early communicative abilities and maternal responses to infant's communicative behaviors in mother-ELGA infants dyads.

2.1.3 Structured assessment methods

Structured assessment methods consist of tests conducted in laboratory, where conditions are the same for all participants. These tools are administered by trained examiners and are characterized by direct and structured examiner-infant interaction. The examiner observes and scores the performance of the infant during tasks that test various skills (e.g., receptive/expressive communication skills). Structured tools provide an index of infant's competences based on presence/absence of behaviors.

One of the most widely used instruments is the Bayley Scales of Infant and Toddler Development, Third Edition (BSID-III, Bayley, 2006). The BSID-III was designed to measure receptive and expressive communication abilities, and also fine and gross motor skills, and cognitive development in TD infants and infants with developmental delays, from 16 days to 42 months and 15 days. It involves a series of face-to-face, structured interactions between the examiner or clinician and the infant, in which a variety of toys, objects and prompts are employed in order to elicit the infant's bids. The BSID-III provides standardized language, motor and cognitive scores, and has been shown to be a valid tool in both research and clinical practice (Sansavini et al., 2014). Recently, it has been using in studies on ELGA preterm infants to assess their language, cognitive, and motor skills at early age (Rahkonen, Heinonen, Pesonen, Lano, Autti, et al., 2014; Sansavini et al., 2014). With regard to the Italian population, an Italian adaptation and

standardization of the BSID-III is available (Ferri, Orsini & Stoppa, 2009; Ferri, Orsini, Rea, Stoppa, & Mascellani, 2015). One advantage of this method regards the direct examiner-child interaction that minimizes the possible variability that caregivers may contribute to the display of infant's early skills. Moreover, differently by the observation of spontaneous behaviors, this tool provides standardized scores that allow the identification of developmental delays. However, by investigating the effectiveness of the BSID-III to track preterm infant's development, some authors (Lobo, Paul, Mackley, Maher, & Galloway, 2013) have highlighted instability of the delays classification in low- and high-risk preterm infants in the first two years of life. Other potential limitations are that involves interaction with an unfamiliar adult in an unfamiliar setting, factors that may impact the performance of at least some infants (Wetherby, 2006).

2.2 Which measures for evaluating the communication development of mother-extremely low gestational age infant (ELGA) dyads at early age?

Each of the methodologies described above provides valuable information about the development of the communicative abilities in young children with and without developmental delay. As I will describe in the next sections, the majority of studies concerned with early communicative development in preterm infant have employed parental report measures. We believe that procedures that use the observation of the spontaneous mother-infant interaction in semistructured or naturalistic setting may provide a more ecological and detailed description of the ELGA infants' communicative repertoires (Crais, Watson, & Baranek, 2009; Tager-Flusberg, Rogers, Cooper, Landa, Lord, et al., 2009) and better highlight early delays. As noted above, observation of the spontaneous mother-infant interaction is also the most appropriate measure to bring out the communicative strategies that the mothers apply with their infants.

Thus, the current research employed the observation of the spontaneous mother-infant

interaction as privileged instrument for detecting the infant and maternal communicative behaviors of interest. We also employed a parental report measure and a structured tool, because we believe that incorporating all these types of measures we can obtain the most complete description of the ELGA infants' developmental level at early age.

3. EARLY COMMUNICATION DEVELOPMENT AND MATERNAL RESPONSES

3.1 Dynamic Systems Theory (DST): a theoretical model of communicative development

The conceptual framework employed in this study is based on the Dynamic Systems Theory (DST; Thelen and Smith, 1994). DST is a recent theoretical approach to the study of development. The term *dynamic systems*, in its most generic form, means systems of elements that change over time. As stated by a main principle of DST, “development can only be understood as the multiple, mutual, and continuous interaction of all the levels of the developing system, from the molecular to the cultural” (Thelen & Smith, 1994). Thus, understanding the progression of communicative development in TD and atypical developing infants, requires investigations of multiple individual components (e.g., gestural and speech modalities, motor skills) and assessment of interactions between components as they unfold over time, considering also the role of environmental factors (Parladé, 2012).

With regard to individual components, in the first years of life they operate collectively and the specific relationships among them are continually adaptable, flexible, and in transition; the communicative system then continually reorganizes while searching for new patterns of stability (Smith & Thelen, 2003). Thus, a myriad of behavioral modes or cooperative coordinations are possible depending on the relative stability of the constituent elements of the system at a given time

(Iverson & Thelen, 1999). An important implication of this view is that instability in one component of the communicative system (e.g., introduction of a new skill or evolution of an existing skill) can translate into varied developmental trajectories; thus, disruption in one component of the system (e.g., motor difficulties) may profoundly alter the way in which all behavioral patterns evolve (Parladé and Iverson, 2011).

An evidence of these intra-individual relationships is the relative degree of temporal coordination between component behaviors (Parladé, 2012). Thus, for example, learning to reach involves the coordination of arm movement and patterns of muscle activation in order to obtain a desired object (Iverson, 2010). Gesture and speech co-occur during production because they are linked to one another and to the same underlying thought processes; moreover, production of gesture-speech combinations in which the two elements are synchronous increases with development (Pizzuto, Capobianco, & Devescovi, 2005).

Recently, some authors have focused on the relationships between gestural, vocal and motor components, highlighting their influence on language development. Infants move through and engage with their surroundings, and these everyday experiences have effects that extend beyond the motor domain to the communicative system (Gentilucci, Santunione, Roy, & Stefanini, 2004; Iverson, 2010). Thus, developmental advances in motor skills (e.g., new actions with hands and mouth) in infancy create a broad range of novel explorative experiences (e.g., object manipulation) and promote the emerging of new communicative opportunities (e.g., use of gestures, improvement of vocal abilities) that may have implications for language development (Iverson, 2010). At the same time, disruptions in the process of motor development should therefore have an effect on gestural and vocal production, and vice versa (Iverson & Thelen, 1999).

Further, the many individual elements of the system are embedded within and open to influence from a complex environment (Smith & Thelen, 2003). DST suggests that these contextual factors and their time functions are the critical aspects of performance and development. For example, the

developmental advances in gestures and the transition from vocalization to babbling change the ways that caregivers react to their infants (Goldstein & West, 1999; Oller, 2000), making the social interaction with the caregiver more effective. Thus, infant and maternal communicative capacities interact to generate the development of more advanced infant behaviors (Thelen & Smith, 1994).

The principles of DST are particularly useful for studying language development, in particular in populations at risk for language delays, such as ELGA infants. In fact, more and stricter associations than dissociations among abilities were found in preterm children, similarly to the trend described in other populations with atypical development (Karmiloff-Smith, 2009). Moreover, ELGA infants appear particularly susceptible to instability in motor skills, which may lead to the delayed or atypical development of communicative abilities. As suggested by Sansavini and colleagues (2011), “preterm birth gives rise to atypical developmental trajectories, characterized by different rates of development and different relationships among competencies; the extent of the deficits varies as a function of the complex interaction among biological and environmental constraints, developmental timing and type of competence, highlighting the dynamic process of development”.

In light of the above, the present research examined the early multimodal communication abilities (gestures, vocal productions, and combinations between them) and motor skills in ELGA and FT infants at an early age, and aimed to verify the reciprocal influences among them and with the caregiver communicative behaviors. Before proceeding to a discussion of the studies 1 and 2, I will describe below what we already know and what would be interesting to study in deep about these topics in the preterm population.

3.2 Communicative abilities of TD infants and preterm infants during the first two years of life

A developmental domain particularly affected in preterm children is language (Barre, Morgan, Doyle, & Anderson, 2011; Sansavini, Guarini, & Caselli, 2011; Van Noort-van der Spek, Franken, & Weisglas-Kuperus, 2012). A small number of longitudinal studies have investigated whether the risk for language impairment is greater among very preterm children (very low gestational age, VLGA, gestational age < 32 weeks) compared to FT children. These studies indicate that, relative to their FT peers, VLGA children exhibited a higher risk for language delay/impairment in the preschool years. Language delay/impairment was exhibited by 30-34% of VLGA children between the ages of 2 and 4 years, but only by 5-10% of FT children (Sansavini, Guarini et al., 2010; Woodward, Moor, Hood, Champion, Foster-Cohen, et al., 2009). In light of this enhanced risk, it is critical to investigate early components of language development that may be informative about possible subsequent language delays in the preterm population.

Early components of language development have been studied extensively in TD infants. The onset of communicative gestures (e.g., Bates, Benigni, Bretherton, Camaioni, & Volterra, 1979; Bavin, Prior, Reilly, Bretherton, Williams, et al., 2008; Capirci & Volterra, 2008), babbling (e.g., Oller, Eilers, Neal, & Schwartz, 1999; Stoel-Gammon, 1989; 2011), and first words (e.g., Caselli, Rinaldi, Stefanini, & Volterra, 2012; Reilly, Bavin, Bretherton, Conway, Eadie, et al., 2009) in the first year of life are signs of the typical course of language development. In addition, at around the end of the first year, TD infants begin to combine gestures and vocal utterances into tightly timed communicative coordinations. Relative to isolated gestures and vocal utterances, these coordinations are more effective in eliciting parental responses and may promote joint attention and language acquisition (Goldin-Meadow et al., 2007; Tamis-LeMonda et al., 2001; Tomasello,

Carpenter, & Liszkowski, 2007).

Less is known about the development of gesture, babbling, first words, and communicative coordinations in infants who are at risk for delayed language development, as are preterm infants. The main studies of gesture and vocal production in this population focused on VLGA infants and generally examined gestures and vocal utterances separately. Regarding gestural production, some studies indicate that relative to FT infants, VLGA infants exhibit slower development of gestural communication at 12 months, as measured via parental questionnaires such as the MacArthur-Bates Communication Developmental Inventory (MB-CDI) (Ortiz-Mantilla et al., 2008; Sansavini, Guarini, Savini, et al., 2011; Stolt, Mäkilä, Matomäki, Lehtonen, Lapinleimu, & Haataja, 2014). However, in an observational study, Suttora and Salerni (2012) did not find significant differences in the number of gestures produced by VLGA and FT infants at 12 months.

Findings on the development of vocalization and babbling in preterm infants have also been somewhat mixed. Research conducted with direct measures (e.g., observation during spontaneous mother-infant play interaction) reported significantly less advanced vocal production in VLGA infants compared to FT infants at 12 months (D'Odorico et al., 2011), and in extremely low birth weight infants (ELBW) relative to FT infants at 8, 9, and 10 months of age (Torola et al., 2012). By contrast, other research using parental questionnaires did not reveal differences between preterm and FT infants in this kind of production (Gonzalez-Gomez & Nazzi, 2012; Stolt, Lehtonen, Haataja, & Lapinleimu, 2012). These contrasting results may be partly due to differences in the methods used to assess the early productions and in sample criteria selection; some authors also suggested that the emergence of speech-like babble may be slightly advanced in these infants, as a result of earlier extrauterine exposure to speech input or intervention (Eilers, Oller, Levine, Basinger, Lynch, & Urbano, 1993).

With regard to word production in preterm infants, studies using the MB-CDI identified differences between VLGA and FT infants in vocabulary size at 16 months (Ortiz-Mantilla et al.,

2008), and at 18 and 24 months (Sansavini, Guarini, & Savini, 2011; Sansavini, Guarini, Savini, et al., 2011; Stolt, Klippi, Launonen, Munck, Lehtonen, et al., 2009), but not at 12 months. D'Odorico and colleagues (2011) replicated this pattern of findings in an observational study conducted during mother-infant play interactions at 18 and 24 months. Furthermore, a recent study employing a lexical task found a lower rate of noun and predicate production in ELGA children at 24 months, compared to FT peers (Sansavini, Bello, Guarini, Savini, Alessandrini, et al., 2015). To our knowledge, however, only one study has shown that differences emerge already at the end of the first year of life in the onset of first words, with delays in ELBW infants with respect to FT infants (Torola et al., 2012).

Relatively little is known about early communicative coordinations in preterm infants. To our knowledge, only two studies to date have investigated the combination of gesture and vocal utterances in VLGA infants. The first study (Suttora & Salerni, 2012) reported that VLGA infants produced fewer gesture-plus-word combinations at 18 and 24 months than did FT infants. The second study (Sansavini, Bello, et al., 2015) found a lower rate of spoken-gestural combinations in a task of predicate production in ELGA children compared to FT peers at 24 months. However, whether such differences exist even earlier in development is not known.

Finally, early communicative abilities seem to have a predictive value on the subsequent language development in preterm populations. Some authors (Sansavini, Guarini, Savini, et al., 2011) studying the action/gesture production and the early word comprehension and production in preterm infants have found an increasing divergence with respect to FT infants in these abilities from 12 to 24 months and a strong predictive value of word comprehension/production at 12 months on word production at 24 months; they also found that action/gesture production at 18 months predicts word production at 24 months of age. Other authors found that receptive and expressive lexicon at 12 months also predicted lexical comprehension and verbal fluency at 3 years both in preterm and FT groups (Rose, Feldman, & Jankowski, 2009). Stolt and colleagues (2009, 2014) found that receptive

lexicon and gestures evaluated at 12 and 15 months using CDI predicted the performance in the Reynell Developmental Language Scales at 24 months (RDLS, Edwards, Fletcher, Garman, Hughes, Letss, & Sinka, 1997; Kortessmaa, Merikoski, Warma, & Varpela, 2001). Thus, in the first year of life, communicative-linguistic abilities that appear affected by preterm birth may result in cascading effects on later development (Sansavini, Guarini, & Caselli, 2011).

Because an accurate and detailed characterization of the communicative abilities at early age may shed light on potential signals of subsequent communicative delays, the present research investigated gestures, vocal behaviors and communicative coordinations at 12 months in ELGA and FT infants. This will provide a more comprehensive indication of the repertoire of behaviors that are involved in early communication and the interrelationships among them, and shed additional light on the extent to which these behaviors may be delayed in ELGA infants.

3.3 Relationship between early communicative abilities and motor skills in TD and preterm infants

The motor domain appears particularly vulnerable in preterm infants (De Kieviet, Piek, Aarnoudse-Moens, & Oosterlaan, 2009) and some of the difficulties found in language achievements of these infants seem to be related to some aspects of motor development (Sansavini, Guarini, & Caselli, 2011). Studies of motor development in TD infants have shown that improvements in control of head, trunk and limbs, which permit the transition from lying to sitting to standing postures (Spencer, Vereijken, Diedrich, & Thelen, 2000), eye-hand coordination with reaching and grasping movements (Rochat & Goubet, 1995; Von Hofsten, 2007), and thumb-fingertip grasp (Jovanovic & Schwarzer, 2011) are signs of the typical course of motor development across the first year of life.

Some studies have found delays in the acquisition of early motor milestones in very preterm

infants (De Kieviet et al., 2009; Jeng, Lau, Hsieh, Luo, Chen, et al., 2008; Van Haastert, de Vries, Helders, & Jongmans, 2006). For example, motor difficulties are evident in VLGA infants with white matter abnormality, who showed gross and fine motor dysfunctions at 12 months (Spittle, Boyd, Inder, & Doyleet, 2009), and also in high-risk preterm infants, who exhibited poorer manipulation skills at 9 months with respect to low-risk preterm and FT infants (Ruff, McCarton, Kurtzberg, & Vaughan, 1984). Motor difficulties have also been observed in VLGA infants without cerebral abnormalities compared to FT infants as early as the first months of life, specifically in head and arm control and in looking behaviors (Van Beek, Hopkins, Hoeksma, & Samsom, 1994). Studies of early motor skills in ELGA infants without major cerebral damage are few in number. In one such study Sansavini, Savini et al. (2011) showed that, relative to VLGA and FT infants, ELGA infants obtained lower locomotor, eye-hand coordination, and cognitive performance scores on the Revised Griffith Mental Development Scales- GMDS-R (Griffiths, 1996) at 6, 12, 18, and 24 months of age. Another study (Sansavini et al., 2014) reported that motor, cognitive, and language scores on the BSID-III were significantly lower for ELGA infants compared to FT infants at 12, 24 and 30 months, and that the divergence between the motor trajectories of the two groups significantly increased over time. The authors hypothesized that these motor difficulties may negatively affect early communicative abilities, such as face-to-face interaction and joint attention, and consequently they may have negative cascading effects on language acquisition in these infants (Sansavini et al., 2014).

Recent evidence points to the existence of a relationship between motor skills and communicative-linguistic abilities in TD infants (Clearfield, 2011; Ejiri & Masataka, 2001; Hill, 2001; Iverson, Hall, Nickel, & Wozniak, 2007; Iverson, 2010; Leonard & Hill, 2014; Zambrana, Ystrom, Schjølberg, & Pons, 2013) and between early motor and communicative delays in infants at risk for developmental concerns (e.g., infants at risk for Autism Spectrum Disorder; Bhat, Galloway, & Landa, 2012; Bhat, Landa, & Galloway, 2011; LeBarton & Iverson, 2013). Given that

motor delays are relatively common in preterm infants (De Kievet et al., 2009; Marlow, Hennessy, Bracewell, & Wolke, 2007; Sansavini et al., 2014), it is surprising that no studies to date have investigated the relationship between early motor development and communicative abilities in preterm infants.

Although early infant communicative and motor skills are often examined separately for practical purposes, it is important to investigate the dynamic and interactive relationships among them to better understand the language development (Iverson, 2010). As a central tenet of DST suggests, complex phenomena (e.g., communication) cannot be fully understood by separating the system by the influences of other individual components (Thelen & Smith, 1994). Because we suppose that delays in motor experiences may constrain learning opportunities (LeBarton & Iverson, 2013) in ELGA infants, in this work we explore the relationships between motor skills and communicative abilities in this clinical population, compared with those of FT infants.

3.4 Maternal responses to TD and preterm infant's communicative behaviors and language development

At 12 months of life TD infants demonstrate a rich and advanced gesture repertoire and start to produce the first intelligible words that enable them to more effectively interact with a social partner (Capirci & Volterra, 2008; Caselli et al., 2012; Tomasello et al., 2007). Two aspects have been examined.

On one side, sensitive responses, i.e. responses focused on infant's requests and interests, that the social partner, caregiver in particular, gives to these infant's communicative signals play a central role in the development of the communication abilities (Wu & Gros-Louis, 2014). In particular, maternal contingent responses (i.e. responses promptly provided) and relevant responses (i.e. responses focused on infant's behaviors and that provide meanings to them) have been shown

to predict later language achievements in TD infants (Baumwell et al., 1997; Girolametto et al., 1999; Goldstein & Schwade, 2008; Gros-Louis et al., 2014; Harris, Jones, Brookes, & Grant, 1986; Rollins, 2003; Tamis-LeMonda et al., 2001). In fact, optimum occasions for language learning occur when adult speech is focused on and relevant to the child's focus of attention (Bloom, 1993, 1998; Tamis-LeMonda et al., 2001).

Concerning maternal contingent and relevant responses, studies on TD infants showed that mothers' ability to respond promptly and in an appropriate manner to the infant's communicative behaviors during the first year of age was positively related with the infant's language development during the second year of life (Gros-Louis et al., 2014; Rollins, 2003). Descriptive expressions, vocal imitations, verbal expansions and reformulations of infant's vocal utterances had a positive effect on infant's lexical and grammatical development (Harris et al., 1986; Girolametto et al., 1999; Tamis-LeMonda et al., 2001), and social feedback to infant's babbling facilitated rapid phonological learning (Goldstein & Schwade, 2008). Moreover, Baumwell and colleagues (1997) found that maternal labeled verbal sensitivity at 9 months predicted infant's language comprehension at 13 months, especially in infants who were initially lower in this ability. This maternal labeling, that is producing a word that could be considered a translation of the infant's gesture or vocal utterance, represents a particularly important environment input for the infant's language development (Goldin-Meadow et al., 2007). Goldin-Meadow and colleagues (2007) demonstrated that when a mother translated her infant's gesture referent into words, those referents were more likely to become part of the infant's word vocabulary than referents that were not translated, and a recent study (Olson & Masur, 2015) found that maternal labeling responses at 13 months predicted the infant's expressive vocabulary at 17 months. By contrast, less contingent and sensitive styles toward the infant's communicative behaviors appear negatively related with the infant's language development (Masur et al., 2005; Paavola et al., 2005).

On the other side, some studies suggested that infants play an important role in shaping their

own environment input. In fact, caregivers were more likely to translate the infant's gestural or vocal behaviors when the infant produced pointing gestures with respect to less developmentally advanced gestures (Kishimoto et al., 2007; Masur, 1982; Olson & Masur, 2011), and to imitate and expand on more advanced nonword vocal utterances (e.g. babbling) than less complex vocalizations (Gros-Louis, West, Goldstein, & King, 2006). These studies suggest that caregivers are more likely to respond with rich communicative input to infants' behaviors that are more developmentally advanced. However, some authors make an alternative hypothesis that less advanced infant's communicative behaviors urge the caregiver to produce contingent and relevant linguistic input. Delays in early spontaneous communication, heightening the levels of concern of the mothers about their infants' development, may make the caregiver particularly vigilant about scaffolding language (Leezenbaum et al., 2014).

Notwithstanding the need to investigate whether early communication difficulties of the extremely preterm infants could shape the way to which the mothers respond to communicative signals of their infants, only few studies, to our knowledge, have analyzed how these mothers respond to the infant's communicative behaviors during the first years of life.

With regard this topic, the majority of studies have focused on mothers of infants born very preterm (very low gestational age, VLGA, gestational age < 32 weeks), and they have reported conflicting results (for reviews, see Bozette, 2007, and Korja, Latva, & Lehtonen, 2012). Some authors (Reissland & Stephenson, 1999) found that mothers of VLGA infants, at 6 weeks after discharge, responded significantly more often to their infants' vocalizations, relative to mothers of FT infants; for these authors this maternal behavior reflected a tendency to impose more structure to the infant's vocalizations to compensate for the lack of participation of their infant. Salerni and colleagues (2007) suggested that mothers of VLGA infants of 6 months of age, relative to mothers of FT infants, often were very active and stimulating during dyadic exchanges to compensate for the lower responsiveness of their infants and to support their engagement. Some authors (Barratt,

Roach, & Leavitt, 1992) found that mothers of preterm infants (gestational age < 34 weeks) displayed sensitive communicative responses toward their infants and that they reinforced their infant's vocalizations more frequently than did the mothers of FT infants at 4 months of age. These authors (Barratt et al., 1992; Salerni et al., 2007) discussed whether this type of behavior supported or was detrimental to the infant's development.

Other studies seem to suggest that the mothers of preterm infants are careful to provide sensitive responses to the infant's communicative cues as the mothers of FT infants do. Some authors (Landry, Smith, Miller-Loncar, & Swank, 1997) who examined the maternal responsiveness of mothers of very low birth weight infants (VLBW), divided into medically high risk (HR) and low risk (LR) infants on the basis of the presence of specific medical complications, did not find differences between HR, LR, and FT groups, from 6 to 40 months of age. Moreover, these authors found that maternal behaviors that were attuned with the infant's developmental level and sensitive to their focus of interest promoted greater increases and faster rate of cognitive and language skills of all groups, than controlling or oversimplified maternal behaviors, with relationships stronger for the HR infants than for the LR and FT infants. Only one study investigated in mothers of extremely low gestational age infants (ELGA, GA < 28 weeks) maternal sensitive-responsiveness toward the infant's communicative behaviors and actions at two years of age, revealing no differences between the mothers of ELGA and FT infants (Rahkonen et al., 2014). These authors found also that maternal sensitive-responsiveness was associated with receptive and expressive language at two years of age. However, we noticed that these studies recruited heterogeneous samples of preterm infants, including LR preterm infants (Barratt et al., 1992) that rarely exhibit early communicative delays, or ELGA infants with white matter abnormalities (Landry et al., 1997; Rahkonen et al., 2014), making difficult the interpretation of these findings. Furthermore, relevancy of maternal responses to the infant's communicative behaviors has not been specifically investigated in the preterm population.

Relevant issues concerning maternal communication with high risk preterm infants, especially healthy ELGA infants, remain thus open. It appears therefore important to run studies that examine whether mothers of ELGA infants differ from mothers of FT infants in the contingency and relevancy of their maternal responses to the infant's communicative behaviors. In particular, there is a need for longitudinal studies that analyses these types of maternal behaviors in relationship to infant's communicative abilities and their effects on the subsequent language development in this clinical population. The present research was designed to address these issues.

4. STUDY 1^{1 2}

Guided by the DST perspective, in the present study we investigated early multimodal communication and the relationship between gestures and motor skills in ELGA infants.

Because of early sub-optimal biomedical and environmental conditions, premature birth represents an event that can have negative impacts on development in multiple domains. For instance, extremely preterm birth is a risk factor for language impairment (Sansavini, Guarini et al., 2010; for review, see Sansavini, Guarini, & Caselli, 2011). Identifying predictors of language development is theoretically and clinically important because it can shed light on potential mechanisms underlying language acquisition in a population at-risk for delay and impairment and suggest potential targets for intervention. It is therefore surprising that relatively little work has addressed this issue in preterm infants.

The literature on TD infants has indicated close relationships between language development and both prelinguistic communicative behaviors (e.g., gesture, vocalization, gesture-vocalization coordinations) and fine motor skills (Hill, 2001; Iverson, 2010; Leonard & Hill, 2014). However, relatively few studies investigated early gestural and vocal abilities in preterm infants and they have

1 Findings of Study 1 have been partially published in Benassi, Savini, Iverson, Guarini, Caselli, Alessandrini, Faldella, & Sansavini (2016), *Research in Developmental Disabilities*, 48, 132-144.

2 This work was supported by grants from the Italian Ministry of Education, University and Research (grant year 2008, protocol 2008J2WEEK, and title: PRIN 2008 “Gestures and language in children with atypical and at risk developmental profiles: relationships among competences, mother-child interaction modalities and proposals of intervention”) and the University of Bologna (grant year 2007, protocol STRAT07SAN, title: Strategic Project 2007 “Early communicative-linguistic and cognitive abilities: risks linked to preterm birth”), awarded to Prof. Alessandra Sansavini.

not generally examined the production of communicative coordinations, which are an important achievement in early communicative development (Iverson, Capirci, Volterra, & Goldin-Meadow, 2008). Moreover, the existing data are conflicting, potentially due to variation in sample selection criteria and tasks used to assess early production (Barre et al., 2011; Marlow et al., 2005).

With regard to the relation between language and fine motor abilities, studies of TD infants have found relationships between increasing refinement in infants' object exploration activities, action imitation, and achievements in language development (Lifter & Bloom, 1989; Zambrana et al., 2013). Thus, delays in motor experiences may constrain learning opportunities (LeBarton & Iverson, 2013) in ways that may impact language development.

4.1 Aims of study 1

The present study had two main goals. The first was to examine the production of communicative behaviors during spontaneous mother-infant play in 12-month-old (corrected age) ELGA infants and compare them to FT infants. Spontaneous gestures (requesting/reaching, pointing, showing, giving, conventional, and representational), vocal utterances (vocalizations, babbling, words), and communicative coordinations (i.e., gesture with gaze, vocalization/babbling with gaze and/or gesture, word with gaze and/or gesture) were examined in detail. We expected that, relative to FT infants, ELGA infants would have smaller repertoires of gestures, vocal utterances and communicative coordinations.

The second goal was to examine motor abilities in ELGA and FT infants using the BSID-III and to explore relationships between communicative behaviors, in particular gestures (which involve motor abilities), and fine and gross motor skills. In order to obtain a complete clinical assessment, cognitive and language skills were assessed using the BSID-III, and the standardized parental report measure PVB-GW was also administered to the parents, in both groups. In line with previous

research (Sansavini et al., 2014), we expected that the ELGA group would exhibit poorer communicative behaviors and fine and gross motor abilities compared with the FT group. Close relationships between gestures and motor skills were also expected. In particular, it was hypothesized that early fine motor difficulties in ELGA infants would reduce opportunities to actively explore objects, gaze to the mother's face, and participate in triadic interactions; thus, they may negatively impact the emergence of communicative behaviors (Leonard & Hill, 2014).

4.2 Method

4.2.1 Participants³

This study involved 40 monolingual Italian infants: 20 ELGA infants and 20 FT infants. The ELGA group (9 males, 11 females) was born at the Neonatal Intensive Care Unit (NICU) of Bologna University. The NICU was accessible to parents day and night and physical contact between parents and their preterm infants in the incubators was encouraged. Cranial ultrasound scans (US) were carried out for all neonates within the first 4 days of life and then repeated weekly during the first month of life. Those neonates with abnormal US in the first month of life were re-examined weekly until normalization, and then two times per month until discharge. After discharge, all preterm infants returned for re-examination with US at the presumed date of birth and again at 3 months of life (corrected age) and entered a medical and neuropsychological follow-up program at the Day-Hospital of the Unit of Neonatology at Bologna University.

Preterm infants were recruited into the present study if they met three primary medical criteria: (a) GA \leq 28 weeks, determined by the date of the mother's last menstrual period and confirmed by first trimester early ultrasonography; (b) no indication of major cerebral damage as detected by ultrasound (US) and confirmed by magnetic resonance imaging at 40 weeks of GA (i.e.,

³ All data were collected by Dr. Silvia Savini at the Unit of Neonatology of the Bologna University.

periventricular leukomalacia -PVL-, intra-ventricular hemorrhage -IVH- > II grade, hydrocephalus) or of congenital malformations; (c) no indication of visual (retinopathy of prematurity -ROP- > II grade) or hearing impairment. Infants included in the sample had some medical complications. These included small for gestational age (SGA, $n = 2$, 10%), respiratory distress syndrome needing mechanical ventilation (RDS-MV, $n = 20$, 100%), bronchopulmonary dysplasia (BPD, $n = 13$, 65%, defined as need of supplemental oxygen at 36 weeks of postconceptional age), IVH of grade I or II ($n = 1$, 5%) detected by US, ROP of grade I or II ($n = 13$, 65%), and hyperbilirubinemia treated with phototherapy ($n = 14$, 70%). In addition, 17 (85%) ELGA infants had persistent hyperechogenicity (HE) of white matter (≥ 14 days), but this had completely resolved by 3 months (for further details, see Table 1).

The ELGA infants had a mean GA of 25.7 weeks ($SD = 1.4$, range = 23-28) and a mean birth weight of 803 grams ($SD = 191$; range = 509-1093). In this group, gestational age and birth weight were highly correlated ($r = .683$). Fifteen (75%) infants were first-born and five (25%) were second- or later-born. All were from families living in the Emilia-Romagna region. The group of ELGA infants was equally distributed across the general range of socioeconomic status (SES), as estimated from mothers' highest level of educational attainment: 12 (60%) mothers had a middle/low educational level (completed high school or at least basic education) and 8 (40%) mothers had a high educational level (completed University/Master's degree). The mean age of mothers was 36.2 years ($SD = 4.8$, range = 27-44). Mothers with a positive family history of psychiatric illness, were excluded.

The comparison group consisted of 20 healthy FT infants (11 males, 9 females) born in the same Hospital as the ELGA group. All FT infants had normal births (GA ≥ 38 weeks and birth weight ≥ 2500 g), and had no history of medical complications (for further details, see Table 1), major cerebral damages, congenital malformations, or visual or hearing impairments. These infants had a mean gestational age of 39.5 weeks ($SD = 1.2$; range = 38-42) and a mean birth weight of

3476 grams ($SD = 464$; range = 2500-4200). Eighteen (90%) FT infants were first-born and two (10%) were second- or later-born. All FT infants were living in the Emilia-Romagna region. Like the ELGA group, these infants' background was equally distributed across SES based on mothers' highest level of education: 8 (40%) mothers had a middle/low educational level and 12 (60%) mothers had a high educational level. Mean age of mothers was 34.6 years ($SD = 3.1$, range = 30-41). The two groups did not differ significantly on gender [$\chi^2 (1, N=40) = 0.40, p = .527$], birth order (Fisher exact test; $p = .407$), maternal age [$t (38) = 1.285, p = .208$] or maternal level of education [$\chi^2 (1, N=40) = 1.60, p = .206$].

The study met ethical guidelines for human subjects protections, including adherence to the legal requirements of the study country, and received formal approval by the local Research Ethical Committee of the University of Bologna. Parents of the ELGA and FT infants gave informed written consent for participation in the study, data analysis, and data publication.

4.2.2 Procedure

The present study is part of a larger longitudinal study on language development in ELGA infants. In this paper, we present data on production of communicative behaviors and motor, cognitive, and linguistic skills in ELGA and FT groups at 12 months of age. As in many studies on preterm infants' development in the first 2 years of life, ELGA infants' age was corrected in order to take into account their level of neuropsychological maturation as assessed via mental and psychomotor scales (Johnson & Marlow, 2006; Pietz, Peter, Graf, Rauterberg-Ruland, Rupp, et al., 2004; Sansavini, Rizzardi, Alessandrini, & Giovanelli, 1996). The mean corrected age of the ELGA infants at the time of evaluation was 12 months and 6 days ($SD = 9$ days) and the mean chronological age of the FT infants was 12 months and 3 days ($SD = 9$ days). This difference was not statistically significant: $t(38) = 0.49; p = .623$.

All ELGA and FT infants were observed in a quiet room designed for neuropsychological

evaluation at the Day-Hospital of the Unit of Neonatology at Bologna University.

Gesture, vocal utterance, and communicative coordination observation. A 30-min mother-infant play session was video-recorded. Infants and mothers sat together on a mat close to a mirrored wall and were videotaped playing with age-appropriate toys (e.g., car, ball, animal toys) and picture books. Mothers were asked to interact and play with their infants as they normally would.

Assessment of motor, cognitive, and language skills. Motor, cognitive and language skills were assessed with the BSID-III. Each of the three scales (motor, cognitive and language) was administered in an approximately 25-minute session by the second author, a neuropsychologist certified on the BSID-III.

The parental report measure PVB-GW was also administered to the mothers. With regard to this measure, there were missing data for one infant (i.e. the mother of one FT infant did not fill in the questionnaire); therefore, we provided the exact n of FT infants (i.e. 19 FT infants) when presenting the analyses.

4.2.3 Coding

All spontaneous communicative behaviors (i.e., gestures, vocal utterances, communicative coordinations) produced during the 30-min mother-infant play interaction session were coded from the videotapes by a trained coder blind to infant group membership using a computer-based video interface system (INTERACT version 9, Mangold International GmbH, 2012) that permitted time-intensive coding of the videotapes. Infant communicative behaviors were considered spontaneous unless they were directly elicited by a mother's request. Thus, they included infant's "apparent imitation" (i.e., an infant's communicative behavior occurring after 5 seconds from the end of the mother's production) and infant's reformulation of mother's productions (see Vihman & McCune, 1994).

Because session length varied slightly among participants ($M = 27.42$, $SD = 5.25$), all frequency variables were converted to rates per 10 min by dividing the total frequency by the length of observation in minutes, then multiplying it by 10.

Gesture. A gesture was considered communicative if it involved clear effort to direct the caregiver's attention (e.g., through use of eye contact, postural shift, repetition, or vocalization; Iverson et al., 2008; Iverson et al., 1994). Deictic, conventional and representational gestures were coded (Capirci, Iverson, Pizzuto, & Volterra, 1996). *Deictic gestures* included *requesting/reaching* (clear extension of the arm with prone or supine open palm or repeated opening/closing of the hand with the aim to request something), *pointing* (clear extension of the index finger toward a proximal or distal object for the purpose of sharing attention or requesting), *showing* (holding up the object toward the partner while making eye contact), and *giving* (extension of the arm with the object in hand and directed toward the hand of another person). *Conventional gestures* were ritualized gestures (e.g., blowing a kiss to someone) or culturally-defined gestures (e.g., NO with the head, HELLO with the hand). *Representational gestures* stand for a specific referent and their primary semantic content does not change with context. Gestures produced with an object (e.g., DRINK, i.e., the infant pretends to drink bringing a cup to his mouth; CAR, i.e., the infant moves a box pretending that it is a car) and without an object (e.g., TELEPHONE, i.e., the infant pretends to phone bringing his hand close to his ear; BIG, i.e., the infant widens his arms pretending to make a big size) were included in this category.

Vocal utterances. Following Paul and Jennings (1992), vocalization/babbling was coded when the utterance was judged by the parent and the examiner to be non-meaningful; it contained, at a minimum, a voiced vocalic element or a voiced syllabic consonant; it was produced with an egressive airstream; and it was judged to be “speech-like” (i.e., cry and vegetative sounds were not included). Utterances that could not be transcribed confidently after listening to them four times were eliminated. Utterances were coded as separate utterances when they were bounded by 1

second of silence on either side, a breath, adult speech, or falling intonation (Paul & Jennings, 1992, adapted). Vocal utterances were classified into one of three mutually exclusive categories (Stoel-Gammon, 1989): *vocalization* (level 1), characterized by utterances composed of a vowel, a syllabic consonant, a consonant-vowel or vowel-consonant sequence in which the consonant was a glide or glottal; *babbling*, which contained at least one consonant-vowel sequence, in which the place and manner feature of the true consonant did not change (level 2), or at least two true consonants differing in place or manner of articulation (level 3). A vocal utterance was coded as a *word* if it resembled an adult word (plausible phonetic shape), was potentially relevant to the ongoing situation (plausible context of use), and met at least 3 of the following 4 criteria: occurred at least 2 times, was phonetically similar to the target, had a specific referent, or was recognized by the caregiver (Vihman & McCune, 1994).

Communicative coordinations. A *communicative coordination* was coded when two (or more) communicative behaviors overlapped temporally with one another (Capirci et al., 1996; Paradé & Iverson, 2015). Temporal co-occurrence was defined as any overlap between co-occurring behaviors; their production need not have been perfectly simultaneous. Three types of communicative coordinations were coded: gestures combined with gaze directed/shifted to mother (e.g., *giving* plus gaze directed to mother); vocalization/babbling combined with gaze directed/shifted to mother and/or with gesture (e.g. vocalization [a] combined with gaze directed to mother and with *requesting*; babbling [kaku] combined with *pointing*); words produced in combinations with gaze directed/shifted to mother and/or with gesture (e.g. [ba ba], onomatopoeic word meaning “dog”, combined with gaze directed to mother; [bu bu] onomatopoeic word meaning “car” combined with CAR, a repeated movement of the hand pretending that it is a car).

4.2.4 Measures

Bayley Scales of Infant and Toddler Development, Third Edition (BSID-III, Bayley, 2006). The BSID-III motor scale includes two subtests. The fine motor subtest examines visual tracking, reaching, object manipulation, grasping, functional hand skills, and responses to tactile information. The gross motor subtest assesses static positioning (i.e., sitting, standing), dynamic movement (i.e., locomotion and coordination), and balance and motor planning. The BSID-III cognitive scale assesses sensorimotor development, exploration and manipulation, object relatedness, concept formation, memory, and cognitive processing. The BSID-III language scale includes two subtests assessing receptive and expressive communication. The receptive communication subtest assesses preverbal and verbal comprehension, social referencing, and receptive vocabulary. The expressive communication subtest assesses preverbal communication (babbling, gesturing, joint referencing, and turn taking) and expressive vocabulary.

The BSID-III provides standardized motor, cognitive and language composite scores, each with a mean of 100 and *SD* of 15. It also provides standardized scaled scores, each with a mean of 10 and *SD* of 3, respectively for the fine motor, gross motor, receptive communication, and expressive communication subtests. Delay on each of the three scales (motor, cognitive, and language) was defined as a standardized score < -1.5 *SD* below the mean (i.e., ≤ 77), which is a common clinical cut-off (Lobo et al., 2013). Behaviors associated with developmental risk are defined in the Appendix B of the BSID-III technical manual (Bayley, 2006). With respect to the motor scale, muscle tone, hand movement, posture and positioning, voluntary movement, and coordination items are considered in identifying infants at developmental risk.

The BSID-III has been shown to be a valid tool in both research and clinical practice; satisfactory reliability and validity have been reported by the authors (Bayley, 2006), with test-retest reliability ranging from .67 to .94, internal consistency coefficients (using the split half

method) of .87–.93, and moderate to high correlations with measures of similar domains. With regard to the Italian population, an Italian translation/adaptation of the BSID-III is available (Ferri et al., 2009) which has been used in research and clinical practice (Sansavini et al., 2014). Since the Italian standardization is not available for infants younger than 12 months and 15 days of age (Ferri et al., 2015), normative values of the original standardization (Bayley, 2006) have been taken as reference for standardized scores, and an Italian FT sample has been included in the present study for purposes of comparison.

Gestures and Words form of the Primo vocabolario del bambino questionnaire (PVB-GW; Caselli & Casadio, 1995). The PVB-GW questionnaire is the Italian version of the Words and Gestures form of the MacArthur-Bates Communicative Development Inventory (CDI-WG; Fenson et al., 2007), designed for infants from 8 to 17 months. The PVB-GW is a reliable and valid parental report measure of language and communication development which has been used extensively in research with FT and preterm infants (Fenson et al., 2007; Ortiz-Mantilla et al., 2008; Sansavini, Guarini, Savini, et al., 2011). PVB-GW consists of yes/no questions that investigate word comprehension, word production, and gesture/action production. It is organized into two parts. Part I consists of a list of 408 lexical items organized into 19 semantic categories (e.g., animal, vehicle, toy, food and drink, clothing, body part). Part II consists of a list of 63 gestures (e.g., pointing, HELLO with the hand) and actions (e.g., bringing a telephone toy close to the ear, throwing of the ball). The parents were requested to fill in the questionnaire checking the words their infant understands or understands and produces and the gesture/action items their infant produces. The PVB-GW was scored according to manualized procedures (Caselli & Casadio, 1995). Three primary measures of communicative and language development were obtained from the PVB-GW: Words Understood, Words Produced, and Total Gestures/Actions.

4.2.5 Reliability

Coding was performed by the first author and by a second trained coder, both of whom were blind to infant group membership. Inter-observer reliability was calculated on 20% of the ELGA and FT dyads. Cohen's kappa calculated to assess inter-coder agreement for categorical decisions was 0.92 for gestures, 0.90 for vocal utterances, and 0.82 for communicative coordinations.

4.2.6 Statistical Analyses

All statistical analyses were carried out using SPSS 21.0 for Windows with alpha = 0.05. Prior to conducting analyses, data were checked for violation of assumptions using the Kolmogorov-Smirnov test. Because distributions for some of the communicative behaviors were non-normal, nonparametric Mann-Whitney tests were conducted to assess potential differences in communicative behaviors (gestures, vocal utterances, and communicative coordinations) between the ELGA and FT groups. Effect sizes (r) for Mann-Whitney U tests were calculated using the formula $r = \frac{Z}{\sqrt{N}}$, where N is the total number of participants in the whole sample); the standard values of r for small, medium, and large effect sizes are 0.1, 0.3, and 0.5 respectively (Field, 2009, p. 550). Chi-square tests were also performed to compare the distributions of infants in the ELGA and FT groups who did versus did not produce communicative behaviors.

With regard to BSID-III, motor, cognitive and language composite scores and fine motor, gross motor, receptive communication, and expressive communication scaled scores were all normally distributed. We therefore conducted a series of ANOVAs to evaluate differences between the ELGA and the FT infants on motor scores (composite score and fine and gross motor scaled scores), composite cognitive score, and language scores (composite score and receptive and expressive scaled scores).

Spearman's correlations were utilized to examine relationships between gestures, fine motor,

and gross motor scaled scores, and cognitive composite scores.

Regarding PVB-GW questionnaire, Words Understood, Words Produced, and Total Gestures/Actions scores were not normally distributed. Thus, Mann-Whitney tests were conducted to assess potential differences between the two groups on these scores.

4.3 Results

4.3.1 *Spontaneous communicative behaviors: gestures, vocal utterances, and communicative coordinations*

Gestures. The mean rates per 10 min of gestures are presented in Table 2. Inspection of these data reveals that in the ELGA group, the most frequently produced gesture was requesting; giving, conventional, and representational gestures were infrequent. Mann-Whitney tests showed that the ELGA infants produced significantly fewer giving and representational gestures compared to the FT infants (see Table 2), whereas there were no significant group differences for requesting/reaching, pointing, showing, and conventional gestures. Chi-square tests revealed that a significantly lower percentage of the ELGA infants produced pointing (60%) and giving (20%) gestures with respect to the FT infants (pointing: 90%; giving: 65%; see Table 2).

Vocal utterances. Mean rates per 10 min of vocal utterances are presented in Table 3. As it is apparent, both ELGA and FT infants mainly produced vocalizations, followed by babbling (see Table 3).

No significant difference was found between the ELGA and FT infants in the rates of production of vocal utterances from each of the categories investigated (see Table 3). However, Chi-square test revealed that a significantly lower percentage of the ELGA infants (35%) produced words relative to the FT infants (70%) (see Table 3).

Communicative coordinations. The mean rates per 10 min of communicative coordinations are

presented in Table 4. In both groups, the most frequently produced communicative coordination type was vocalization/babbling combined with gesture and/or gaze directed/shifted to the mother (see Table 4). There were no significant group differences in the rate or presence of communicative coordinations.

4.3.2 Motor, cognitive, and language skills on BSID-III

Descriptive data from the motor, cognitive and language BSID-III scales and statistical comparisons using ANOVAs are presented in Table 5.

Relative to their FT peers, the ELGA infants obtained significantly lower composite motor scores (see Table 5). Analysis of the sub-tests scaled scores revealed that both fine and gross motor scores were significantly lower for ELGA infants compared to FT infants (see Table 5). Scores on the cognitive scale also differed significantly, with the ELGA infants performing more poorly than their FT peers (see Table 5). However, no significant differences were found between the two groups on the language scale and the receptive and expressive subtests.

Examination of the numbers of infants who exhibited delays (defined as $< -1.5 SD$ below the mean) on the BSID-III revealed that among the ELGA infants, 4 (20%) had a motor delay (among them one had also a language delay) and one (5%) had a cognitive delay; among the FT infants, one (5%) had a motor delay. However, these differences were not statistically reliable. A descriptive examination of the communicative behaviors of the ELGA infants with motor or cognitive delay indicated that they did not produce any of the following communicative behaviors: pointing, giving, representational gestures, or words.

4.3.3 Communicative-linguistic abilities on PVB-GW

Descriptive data from the PVB-GW (Words Understood, Words Produced, and Total Gestures/Actions scores) and statistical comparisons using Mann-Whitney tests are presented in Table 6.

Mann-Whitney tests showed that the ELGA infants obtained significantly lower Total Gestures/Actions score, relative to their FT peers (see Table 6). There were no significant group differences for Words Understood and Words Produced.

4.3.4 Relationships between gestures, motor and cognitive skills

Finally, correlations between gestures, motor, and cognitive skills and are presented in Table 7. There were significant associations between fine motor scaled scores and pointing and representational gestures in the ELGA group, and between fine motor scaled scores and giving in the FT group (see Table 7). No significant correlations between gross motor scaled scores and gestures were found for either group, except for an isolated negative correlation with conventional gestures in the FT group. Significant associations were also found between the composite cognitive score and representational gestures in the ELGA group, and between the cognitive composite score and conventional gestures in the FT group (see Table 7).

4.4 Discussion

This study shows for the first time, via the combined use of detailed coding schemes, a clinical tool, and a parental report measure, a slower development of gestures and words as well as of motor and cognitive development in ELGA infants at 12 months of corrected age. This slower communicative development was mainly associated with slower motor development.

4.4.1 Differences in communicative behaviors between ELGA and FT infants

The first major finding of this study was that the gesture development of ELGA infants was less advanced than that of FT infants. This less advanced gestural development in ELGA infants was highlighted both using the observation of spontaneous gestural behaviors and the parental report

measure PVB-GW. This result is consistent with evidence suggesting a weakness in gesture production, as indicated by some studies conducted on VLGA infants at 12 months using the MB-CDI (Ortiz-Mantilla et al., 2008; Sansavini, Guarini, Savini, et al., 2011; Stolt et al., 2014). The novel contribution of the present study is the detailed description of the typology and frequency of gestures spontaneously produced by ELGA relative to FT infants. Specifically, deictic gestures that involve sharing an object/interest with a partner, were produced significantly less often by ELGA relative to FT infants. This was the case for the giving gesture, which was less frequent in ELGA infants than in FT infants, and for the pointing gesture, which was present in a significantly lower percentage of ELGA infants. These findings support our hypothesis of a delay at 12 months in ELGA infants in the onset of these important gestural precursors of language development (Bavin et al., 2008; Capirci & Volterra, 2008; Iverson & Goldin-Meadow, 2005), which in TD infants begin to appear at around 9-10 months of age (Caselli et al., 2012; Lock, Young, Service, & Chandler, 1990; Sansavini, Bello, Guarini, Savini, Stefanini, & Caselli, 2010). Representational gestures, which constitute a means for the transition to the use of symbolic communication (Capirci & Volterra, 2008) were also less frequently produced in the ELGA than in the FT group, suggesting that ELGA infants are just beginning meaning construction and sharing at 12 months. A lesser use of representational gestures was also found in a recent study employing a predicate production task which showed that at 24 months ELGA children, differently from FT peers, seldom use representational gestures to express meaning, for predicates which they do not master yet (Sansavini et al., 2015).

With regard to vocal production, our hypothesis was only partially confirmed. In line with other studies in the literature that did not find differences between preterm and FT infants in the first year of life in vocalization and babbling production (Stolt et al., 2012) and in word production (D'Odorico et al., 2011; Sansavini, Guarini, Savini, et al., 2011), we did not find differences between the two groups in the frequency of spontaneous vocalization, babbling, or words at 12

months of age. This result appears in line with the results that we obtained using BSID-III and PVB-GW. In fact, we did not find significant differences between the two groups neither on language BSID-III scores (composite score, receptive and expressive scores) nor on PVB-GW scores (Words Understood and Words Produced). Therefore, the linguistic abilities of the ELGA infants (both expressive and receptive abilities) considered as a group appears at the same level of their FT peers, at least at 12 months of corrected age. As Oller and colleagues (1994) suggest, interpretation of these findings might depend on the recognition that ELGA infants, examined at their corrected age, have a longer environmental experience than FT infants. Although no differences between the two groups emerged in the frequency of vocal behaviors, in the case of spontaneous words we found that, relative to FT infants, a significantly lower percentage of ELGA infants produced words at 12 months during the video-recorded mother-infant interaction session. Given that, in typical development, the onset of first words at around 12 months of life is a positive prognostic sign for language development (Reilly et al., 2009; Sansavini, Bello, et al., 2010), we hypothesize that the delay found in a majority of the ELGA infants in the onset of words may index in a subsequent language delay. Although linguistic difficulties were not detectable at this age with a clinical tool and a parental questionnaire assessing receptive and expressive language, they were apparent in the analysis of spontaneous communicative behavior, indicating the importance of integrating structured tools and parental report measures with observational coding schemes.

Finally, this research examined spontaneous communicative coordinations of gesture and gaze, vocalization/babbling and gaze or gesture, words and gaze or gesture at 12 months for the first time in the ELGA population. From our results it appears that at this age, the ELGA and FT infants do not differ in these types of behaviors. This may be the result of the young age of the infants observed in our study. Thus, the question of whether a delay in the spontaneous communicative coordinations in ELGA infants is observable as early as 12 months remains open. Future research is needed to examine communicative coordination production in ELGA infants at later ages.

4.4.2 Relationships between gestures and fine motor skills in ELGA infants

With regard to the second aim of the present study, the ELGA infants in our group lagged significantly behind their FT peers in both gross and fine motor skills. This result is in line with the few studies that have recently started to investigate the motor abilities of ELGA preterm infants during the first years of life (Sansavini, Savini et al., 2011; Sansavini et al., 2014), and thus contributes to this emerging research landscape. Furthermore, 20% of the ELGA group exhibited a motor delay, while only 5% presented a cognitive or linguistic delay, suggesting that the motor domain may be most affected by an extremely preterm birth at this age. Nevertheless, less advanced development was also observed in ELGA infants in cognitive skills, but not in linguistic skills as assessed on the BSID-III, as mentioned above. These findings suggest that at 12 months corrected age, ELGA infants show apparent difficulties, relative to FT infants, in motor exploration, knowledge, and representation of the world.

A particularly novel and interesting finding from this study has to do with the relationships between fine motor skills and gestures observed in both groups. This result is consistent with a growing body of work indicating that the motor and language domains are closely linked in the brain, particularly in their early developmental stages (Iverson & Thelen, 1999; McNeill, 2005; Zukow-Goldring, 2005). Neurophysiologic evidence on the functioning of the motor system (Arbib, 2005; Rizzolatti & Arbib, 1998) supports the hypothesis of a tight link between motor programs associated with actions, gestures, and spoken linguistic representations (Bernardis & Gentilucci, 2006; Capirci, Caselli & De Angelis, 2010). Notwithstanding, some differences were found between the two groups. In the ELGA group, fine motor scores were positively correlated with pointing and representational gestures, while in the FT group, an association between giving and fine motor skills was found. The associations found in the ELGA group highlight the critical role of fine motor skills for the development of pointing, which is one of the primary early indices of language delay (Bavin et al., 2008; Caselli et al., 2012; Reilly et al., 2009; Stolt et al., 2014), and of representational

gestures, which are the expression of the transition from action to language (Capirci & Volterra, 2008; Capone, 2007). An association between cognitive skills and representational gestures was also found in ELGA infants, highlighting the interaction between motor, cognitive, and communicative skills in the ELGA infants that has also been observed in other populations with atypical development (Leonard & Hill, 2014). This finding brings evidence in favor of the hypothesis that associations among domains may be more evident in populations with developmental delays (Karmiloff-Smith, 2009). As some authors suggest (Bhat et al., 2012; Gernsbacher, Sauer, Geye, Schweigert, & Goldsmith, 2008; Iverson, 2010), slowed or uncoordinated arm, facial, and articulation movements may limit effective head turning, use of gestures, and vocal utterances. Interestingly, the ELGA infants who exhibited a motor or cognitive delay did not produce pointing, giving, representational gestures or first words.

In the light of these findings, we propose that a developmentally important linkage exists between motor, cognitive, and communication delays in ELGA infants. During early development, it is possible that a relatively small disruption in one of the interacting systems (i.e., the motor system) could have negative escalating effects on other systems related to motor development, such as the language domain (Iverson, 2010; Libertus & Needham, 2011).

The implications of these findings for developmental research and clinical applications will be considered in the General Discussion.

5. STUDY 2⁴

With the aim to identify early risk conditions for the ELGA infants' language development, it appears needful not only to study the early communicative abilities of these infants as we have done in Study 1, but also to study environmental factors that may be related to communicative development of ELGA infants (Sansavini, Guarini, & Caselli, 2011). Thus, in the present study, we are interested to investigate specific characteristics of the communicative input that these infants receive by their mothers during the first year of life. The transactional model asserts that infants learn through a dyadic process that involves both their own behavior and the input they receive from adults around them (Sameroff, 2009). From this prospective, infant's and mother's behaviors are mutually influential and this dynamic interaction influences infant's development in significant ways. Thus, verifying whether extremely preterm birth could affect the maternal communicative strategies appears an important goal to better explain the developmental outcomes of these infants. Specifically, we focused on the quality of the maternal communicative responses provided to infant's communicative behaviors at 12 months. To our knowledge, few studies have examined in this clinical population the characteristics of the maternal communicative responses to the infant's communicative signals and there is a dearth of research assessing their relationship to the infant's

4 This work was supported by grants from the Italian Ministry of Education, University and Research (grant year 2008, protocol 2008J2WEEK, and title: PRIN 2008 "Gestures and language in children with atypical and at risk developmental profiles: relationships among competences, mother-child interaction modalities and proposals of intervention") and the University of Bologna (grant year 2007, protocol STRAT07SAN, title: Strategic Project 2007 "Early communicative-linguistic and cognitive abilities: risks linked to preterm birth"), awarded to Prof. Alessandra Sansavini.

communicative behaviors and their predictive value on the infant's language development.

Shedding light on these maternal communicative behaviors and on their influences on the infant's communicative-linguistic skills is an important theoretical and clinical purpose for understanding the potential mechanisms underlying language acquisition in a population at risk for language delay and to suggest potential targets for intervention.

5.1 Aims of study 2

The present study had three main goals. The first goal was to examine contingency and relevancy of the maternal communicative responses to ELGA infants' communicative behaviors measured during spontaneous mother-ELGA infant interaction at 12 months (corrected age) and compare them to the responses of mothers of FT infants. Maternal responses to infant's communicative behaviors were distinguished in maternal non-contingent responses, i.e. responses which occurred later than 5 sec from the end of infant's communicative production (Baumwell et al., 1997), versus maternal contingent responses. Maternal contingent responses were classified in function of their relevancy, i.e. their pertinence to infant's communicative behaviors and informativeness. Thus, a maternal response not focused on infant's communicative behavior or not adding information to the infant's message was considered as a non-relevant response. In addition, the maternal relevant responses were classified into categories, in function of their grade of relevancy, i.e. based on how many labels, that translate the infant inferred communicative intent, the maternal response contained. Thus, relevant responses with no label, relevant responses with only one label, and relevant responses with more labels were distinguished.

In this study, we speculated that maternal responses composed of repeated labeling could support the infant's language development more than responses without label or composed of only one label. The first question addressed in the present study was whether mothers of ELGA and FT

infants differed in contingency and relevancy of responses to their infants' communicative behaviors. Some studies conducted on extremely preterm infants revealed less advanced communicative gestural and vocal productions in the first year of life (see Study 1; Torola et al., 2012). Specifically, as we found in Study 1 of the present research, ELGA infants, relative to FT infants, produced fewer instances of more developmentally advanced communicative gestures (e.g. pointing, giving, representational gestures) as well as fewer words at 12 months. Extremely low birth weight infants (ELBW), compared to FT infants, showed significantly less advanced vocalizations from 8 to 10 months and appeared delayed in the onset of the first words at 12 months (Torola et al., 2012). Based on these findings, in this study we conducted additional qualitative analyses to confirm the hypothesis of less advanced communicative abilities in ELGA infants, with respect to FT infants. Specifically, deictic gestures investigated in Study 1 of the present research, in this study, were merged into two categories, *requesting/showing* and *pointing/giving*, based on evidence that pointing/giving gestures particularly involve sharing attention with the social partner for having a communicative reciprocal exchange, and thus they appear developmentally more advanced (Iverson et al., 1994). This is confirmed by the fact that they tend to appear a bit later in development (Caselli et al., 2012; Sansavini, Bello et al., 2010) than requesting and showing gestures. For the same reasons, the vocal utterances investigated in Study 1, were merged into two categories, *nonword vocal utterances* and *words*, based on evidence that vocalizations and babbling appear early in development and are less advanced than words (Majorano & D'Odorico, 2011). We postulated less advanced gestures and vocal utterances in ELGA infants, compared with FT infants. Consequently, two possible types of maternal communicative behaviors might be expected. The first hypothesis was that, ELGA infants, because of their communication difficulties, afforded their mothers with fewer opportunities to provide contingent and relevant responses to their communicative behaviors. The second hypothesis was that mothers of ELGA infants, being concerned about their infant's communication development, might be attuned with it, and thus

provide contingent and relevant responses as the mothers of FT infants do. In face of communicative difficulties of their infants, these mothers might pay particular attention to provide repeated labeling in response to the gestures and vocal utterances of their infants, similarly to the mothers of FT infants.

The second goal was to explore the relationships between maternal communicative responses and infant's communicative-linguistic abilities at 12 and 24 months (corrected age for ELGA infants). Specifically, we examined the relationship between the grade of relevancy of the maternal responses observed at 12 months and: a) the infant's spontaneous communicative gestural (i.e. requesting/showing, pointing/giving, conventional gestures, representational gestures) and vocal behaviors (nonword vocal utterances and words) at 12 months; b) the infant's receptive and expressive communication abilities, evaluated at 12 and 24 months, using the BSID-III. The second question addressed in this study was whether the grade of relevancy of the maternal responses was related to the infant's communicative and language abilities especially in the ELGA dyads. We hypothesized that the maternal relevant responses characterized by more labels, with respect to those without label or with only one label, would show positive and stricter relationships with the infant's communicative and language abilities, both at 12 months and 24 months, and that these relationships are stricter in the ELGA dyads, relative to the FT dyads, at both ages. According to our hypothesis, the reciprocal influences between infant's communicative productions and mother's communicative responses might be stricter in mother-ELGA infant dyads than in FT dyads at 12 months, being ELGA infants more vulnerable in their development and therefore more susceptible to environmental experience. Moreover, maternal relevant responses might be particularly important at 12 months of age, for ELGA infants. In particular, environmental linguistic input characterized by repeated labeling might affect the ELGA infants' subsequent language skills at 24 months.

Thus, the third goal of the present study was to analyze whether maternal relevant responses

with more labels were predictive of expressive language at 24 months. To our knowledge, none has considered the effect that this specific type of maternal response could have on the subsequent language development of ELGA infants. We speculated that the maternal repeated labeling could have an important effect on the infant's expressive communication abilities, and thus have a supportive role for language acquisition of ELGA infants. The regression model took also into account neonatal status (being ELGA versus FT) and BSID-III expressive communication scaled score at 12 months, in order to understand whether the maternal repeated labeling had a more important predictive value than to be born extremely preterm and/or the expressive communication level at 12 months.

5.2 Method

5.2.1 Participants⁵

Forty mother-infant dyads (20 ELGA and 20 FT dyads) were recruited from the Unit of Neonatology of Bologna University. They were those recruited in Study 1 (see Table 1). Maternal responses were examined when the infants were 12 months old (corrected age for ELGA infants). Two mother-FT infant dyads (one female and one male) did not take part in the second evaluation at 24 months of age (i.e. the mother of the first infant missed the appointment and the mother of the second infant did not come because she was in motherhood); thus, at the second observation, the comparison group consisted of 18 mother-FT infant dyads.

For the detailed description of the participants to Study 2, see Study 1 (par. 4.2.1 and Table 1).

5.2.2 Procedure

The mothers agreed to participate in a longitudinal study that followed their infants from birth

⁵ All data were collected by Dr. Silvia Savini at the Unit of Neonatology of the Bologna University.

until preschool age. In this paper, we present data on the production of maternal communicative responses at 12 months in relationships to infants' communicative and linguistic skills evaluated at 12 and 24 months of age. As in many studies on preterm infants' development in the first 2 years of life, ELGA infants' age was corrected in order to take into account their level of neuropsychological maturation as assessed via mental and psychomotor scales (Johnson & Marlow, 2006; Pietz et al., 2004; Sansavini et al., 1996). At the first time of evaluation, the mean corrected age of the ELGA infants was 12 months and 6 days ($SD = 9$ days) and the mean chronological age of the FT infants was 12 months and 3 days ($SD = 9$ days). This difference was not statistically significant: $t(38) = 0.49$; $p = .623$ (see Study 1, par. 4.2.2). At the second observation, the mean corrected age of the ELGA infants was 24 months and 8 days ($SD = 11$ days) and the mean chronological age of the FT infants was 24 months and 16 days ($SD = 14$ days). This difference was not statistically significant: $t(36) = -1.998$; $p = .053$.

All mother-infant dyads were observed in a quiet room designed for observation and neuropsychological evaluation at the Day-Hospital of the Unit of Neonatology at Bologna University.

Maternal communicative responses to infant's gestures and vocal utterances at 12 months. A 30-min mother-infant play session was video-recorded. Mothers and infants sat together on a mat close to a mirrored wall and were videotaped playing with age-appropriate toys (e.g., car, ball, animal toys) and picture books. Mothers were asked to interact and play with their infants as they normally would.

Assessment of language, cognitive and motor skills at 12 and 24 months. Assessment of language, cognitive and motor skills was done with the BSID-III. For details about this assessment at 12 months, see Study 1 (par. 4.2.2).

With regard to language assessment at 24 months, the BSID-III receptive communication subtest evaluates comprehension of words and simple directions, and the BSID-III expressive

communication subtest examines word production, word-gesture and word-word combinations.

The BSID-III cognitive scale, at 24 months, evaluates concept formation, memory, cognitive processing, and relational play. The BSID-III fine motor subtest, at 24 months, examines functional hand skills, thumb-fingertip, palmar and transitional grasp, imitation of actions with objects and crayons; the BSID-III gross motor subtest evaluates walking forward/backward, walking up/down stairs, throwing a ball and running with coordination.

The BSID-III provides standardized language, cognitive and motor composite scores and also standardized scaled scores for the receptive and expressive communication subtests, and for the fine and gross motor subtests.

As described in Study 1 of the present research, an Italian translation/adaptation of the BSID-III is available (Ferri et al., 2009) which has been using in research and clinical practice (Sansavini et al., 2014). Since the Italian standardization is not available for infants younger than 12 months and 15 days of age (Ferri et al., 2015), normative values of the original standardization (Bayley, 2006) have been taken as reference for standardized scores both at 12 and 24 months of age, and an Italian FT sample has been included at both ages for purposes of comparison.

5.2.3 Coding

With the purpose to code the maternal communicative responses to the infant's communicative signals, we coded all verbal and gestural maternal communicative responses to the infant's gestural and vocal behaviors.

Infant's communicative behaviors. With regard to infants' gestures and vocal utterances, we used the coding carried out in the Study 1 of the present research (see Study 1, par. 4.2.3, for a more detailed coding description).

Maternal communicative responses. We coded only the first maternal utterance that followed the infant's communicative behavior (gesture, vocal utterance, or combination of them). Criteria for

delineating among utterances involved both timing and content: if 2 seconds elapsed between utterances or if the maternal utterance changed of meaning, we stopped to code (Baumwell et al., 1997).

Maternal responses were classified as non-contingent and contingent responses. A *non-contingent response* was coded if it occurred later than 5 sec from the end of the infant's communicative production. A *contingent response* was coded if it occurred within 5 sec from the end of infant's communicative production (Baumwell et al., 1997).

Among contingent responses, non-relevant and relevant responses were distinguished. A *contingent non-relevant response* was coded when the maternal response did not refer to the infant's communicative behavior (e.g. the infant produced a requesting toward an object and the mother said "Look at!" referring to another object) or if it did not provide linguistic information (e.g. the infant produced a pointing toward an object and the mother said "Eh?" or "Mm"). On the contrary, when a maternal response was related to the infant's communicative behavior and provided information, we coded it as *contingent relevant responses* (Girolametto et al., 1999; Tamis-LeMonda et al., 2001).

The contingent relevant responses were distinguished in three categories, according to their grade of relevancy, i.e. in function of the number of verbal labels that the mother produces. A label is a word that can be considered a translation of the infant's gesture or vocal utterance (Goldin-Meadow et al., 2007). A *contingent relevant response with no label* was coded when the mother produced a gesture (e.g. the giving gesture in response to the infant's requesting gesture) or a verbal response that did not contain a verbal label relating to the infant's communicative behavior (e.g. after a vocalization of the infant, the mother said "Nice! Do you sing?"). A *contingent relevant response with one label* was coded if the maternal verbal response contained a single label relating to the infant's communicative behavior (e.g. after a pointing of the infant to a dog toy, the mother said "Yes, the doggy", or after a vocalization concerning the ball, she said "Do you want the ball?"). A *contingent relevant response with more labels* was coded when the mother responded to the

infant's communicative behavior with more than one verbal labeling (e.g. the mother, referring to a toy car that the infant pointed at, said "The car, nice that car!"). Given that, in the latter type of maternal response, the mother repeated more than one time the referent word of the infant's communicative behavior, it was characterized by a higher grade of relevancy, compared with contingent relevant responses without or with one label.

All maternal and infants' communicative behaviors were coded off-line from the videotapes by a trained coder blind to group membership using a computer-based video interface system (INTERACT version 9, Mangold International GmbH, 2012) that permitted time-intensive coding, frame by frame, of the videotapes.

5.2.4 Reliability

Coding was performed by the writer and by a second trained coder, both of whom were blind to mother's and infant's group membership. Inter-observer reliability was calculated on 20% of the ELGA and FT dyads interactions. With regard to the infant's communicative behaviors, Cohen's kappa calculated to assess inter-coder agreement for categorical decisions was 0.92 for gestures, and 0.90 for vocal utterances. Regarding maternal communicative responses, the inter-observer reliability was 0.91.

5.2.5 Statistical Analyses

All statistical analyses were carried out using SPSS 23.0 for Windows with an alpha level of 0.05. Prior to conducting analyses, data were checked for violation of assumptions using the Kolmogorov-Smirnov test and the Levene test. Since the distributions for some of the infant's communicative behaviors were not normal, Mann-Whitney tests were conducted to assess potential differences in communicative gestural behaviors (requesting/showing, pointing/giving, conventional gestures, representational gestures) and vocal behaviors (nonword vocal utterances

and words) between the ELGA and FT groups at 12 months. In few instances, the infants combined gestures with vocal utterances. In these cases, gestures and vocal utterances were separately considered. Effect sizes (r) for Mann-Whitney U tests were calculated using the formula $r = \frac{Z}{\sqrt{N}}$ where N is the total number of participants in the whole sample; the standard values of r for small, medium, and large effect sizes are 0.1, 0.3, and 0.5 respectively (Field, 2009, p. 550).

All types of maternal communicative response were normally distributed; thus ANOVAs were conducted to assess potential differences in the maternal communicative responses (non-contingent responses, contingent non-relevant responses, contingent relevant responses *-with no label, with one label, with more labels*) between the mothers of the ELGA and FT groups at 12 months.

BSID-III language, cognitive and motor composite scores, receptive/expressive communication scaled scores and fine/gross motor scaled scores were normally distributed, both at 12 and 24 months. Thus, analyses of variance (ANOVAs) were conducted to evaluate potential differences on language scores (composite scores, receptive and expressive scaled scores), cognitive composite scores, and motor scores (composite scores, fine and gross motor scaled scores) between the ELGA and the FT infants, at 12 and 24 months.

With regard to the second aim, Spearman's correlations were utilized to examine relationships between maternal relevant responses (i.e. with no label, with one label, with more labels) and infant's spontaneous communicative gestural (i.e. requesting/showing, pointing/giving, conventional gestures, and representational gestures) and vocal behaviors (nonword vocal utterances and words) at 12 month. Pearson's correlations were utilized to examine relationships between maternal relevant responses, BSID-III receptive and expressive scaled scores, and cognitive composite scores, at 12 and 24 months.

Regarding the third aim, a linear regression analysis (backward method) was performed in order to evaluate whether the maternal repeated labeling at 12 months together with the neonatal status (being ELGA versus FT) and the infants' BSID-III expressive communication scaled score at 12

months predicted the BSID-III expressive communication scaled score at 24 months. In the regression analysis, maternal repeated labeling at 12 months, neonatal status (being ELGA versus FT), and infants' BSID-III expressive communication scaled score at 12 months were entered together as independent variables.

5.3 Results

5.3.1 Maternal responses to infant's communicative behaviors at 12 months

Prior to conduct the main analyses on the maternal communicative responses, we investigated infants' spontaneous communicative behaviors by tabulating the frequencies of gestures (i.e. requesting/showing, pointing/giving, conventional gestures, representational gestures), and vocal utterances (nonword vocal utterances and words). Because session length varied slightly among participants [ELGA group: $M = 25.96$, $SD = 6.67$; FT group: $M = 28.87$, $SD = 2.77$; $t(38) = -1.801$, $p = .084$], all frequency variables were converted to rates per 10 min by dividing the total frequency by the length of observation in minutes, then multiplying it by 10.

The mean rates per 10 min of *gestures* are presented in Table 8. Mann-Whitney tests showed that the ELGA infants produced significantly more requesting/showing gestures, but fewer pointing/giving gestures and representational gestures, compared to FT infants (see Table 8). There were no significant group differences for conventional gestures.

With regard to *vocal utterances*, mean rates per 10 min are presented in Table 8. Both ELGA and FT infants mainly produced nonword vocal utterances. Using Mann-Whitney tests, no significant difference was found between the ELGA and FT infants in the rates of production of nonword vocal utterances and words (see Table 8).

Descriptive data for the maternal responses to infant's communicative behaviors and statistical comparisons using ANOVAs are presented in Table 9. Because infants varied in the number of

communicative behaviors they produced, mothers' opportunities to respond to infants also varied. Thus, percentages were utilized to compare the maternal communicative responses of the two groups. Percentages were calculated for all types of maternal communicative responses: non-contingent responses, contingent non-relevant responses, contingent relevant responses with no label, contingent relevant responses with one label, and contingent relevant responses with more labels. This was done by dividing the number of maternal responses (e.g. non-contingent responses to infant's communicative behaviors) by the total number of infant's communicative behaviors (gestures and vocal utterances), then multiplying it by 100.

Descriptive analyses showed that in both groups non-contingent responses provided to the infant's communicative behaviors were about 30% in both groups (31% in the ELGA group and 34% in the FT group). No significant differences were found between the mothers of ELGA infants and the mothers of FT infants in the percentages of non-contingent responses (see Table 9).

With regard to maternal contingent responses, the inspection of descriptive data reveals that in both groups non-relevant responses were about 10% (11% in the ELGA group and 8% in the FT group) with no significant differences between the two groups (see Table 9).

Regarding maternal relevant responses, in both groups, relevant responses with no label were the most frequent ones (39% in the ELGA group and 37% in the FT group), followed by relevant responses with one label (12% in the ELGA group and 13% in the FT group), and by relevant responses with more labels (7% in the ELGA group and 8% in the FT group). None of these group comparisons were statistically significant (see Table 9).

With regard to language, cognitive and motor BSID-III scales at 12 months, this study utilized the data obtained and described above in Study 1 (see Study 1, par. 4.3.2 and Table 5).

Descriptive information concerning language, cognitive and motor BSID-III scales at 24 months is shown in Table 10. Scores on the language scale (composite scores) differed significantly, with the ELGA infants performing more poorly than the FT infants; however, no significant

differences emerged between the two groups on the receptive and expressive communication subtests scaled scores (see Table 10).

Differences were also found between the two groups on the cognitive and motor scales at 24 months, with significantly lower cognitive composite scores, motor composite scores, and fine and gross motor subtests scaled scores in the ELGA infants, compared with the FT infants (see Table 10).

5.3.2 Relationships between maternal contingent relevant responses and infant's communicative-linguistic skills at 12 and 24 months

Correlations between types of maternal contingent relevant responses and infant's spontaneous gestures and vocal utterances at 12 months are presented in Table 11. In the group of the ELGA dyads, contingent relevant responses with no label were negatively correlated with words ($r_s = -.515, p = .020$), whereas maternal responses with more labels were positively related to pointing/giving gestures ($r_s = .503, p = .024$) and words ($r_s = .475, p = .034$). In the FT dyads contingent relevant responses with more labels were positively associated with conventional gestures ($r_s = .554, p = .011$).

Correlations between types of maternal relevant responses and BSID-III language and cognitive skills at 12 months are presented in Table 12. In the ELGA dyads, two significant positive associations emerged between contingent relevant responses with one label and expressive scaled scores ($r_s = .507, p = .023$), and between contingent relevant responses with more labels and receptive scaled scores ($r_s = .602, p = .005$) (see Table 12). No significant correlations between types of relevant responses and receptive/expressive scaled scores were found in the FT group (Table 12). No significant associations were found between types of relevant responses and composite cognitive scores in the two groups (Table 12).

Pearson's correlations showed that, in the ELGA dyads, contingent relevant responses with

more labels at 12 months were positively related to expressive scaled scores at 24 months ($r_s = .542, p = .014$; see Table 13). In both groups, no other significant correlations were found between the other maternal relevant responses at 12 months and the BSID-III scores at 24 months (Table 13).

5.3.3 Predictive effect of maternal repeated labeling on the infant's expressive language at 24 months

The linear regression analysis (backward method), including the maternal relevant responses with more labels at 12 months, the neonatal status (extremely preterm birth versus at term birth) and BSID-III expressive communication scaled score at 12 months as independent variables, showed a final model characterized by an adjusted R^2 of 0.12 [$F(1,37) = 5.994, p = .019$]. Neonatal status and BSID-III expressive communication scaled score at 12 months were removed from the final model, because they were not significant. The maternal relevant responses with more labels variable was a significant predictor of the BSID-III expressive communication scaled score at 24 months (see Table 14).

Such findings can be interpreted as showing that when the other covariates were held fixed, the mean value of the BSID-III expressive communication scaled score at 24 months increased of 0.18 points for a one unit increase in the percentage of maternal relevant responses with more labels at 12 months (see Table 14). Therefore, the unique significant predictor was the maternal relevant responses with more labels ($\beta = 0.38$; see Table 14).

5.4 Discussion

This study has examined, for the first time in mothers of healthy ELGA infants, contingency and relevancy of the maternal responses provided to the infants' communicative behaviors at 12 months of age, compared to mother-FT infant dyads, in the theoretical perspective of considering

mutually influential the caregiver's and infant's communicative behaviors (Sameroff, 2009). Strict relationships between maternal relevant responses and infant's communication abilities at 12 months, and a predictive effect of the maternal repeated labeling responses on the infant's linguistic skills at 24 months in ELGA dyads, were found.

5.4.1 Contingent and relevant maternal responses to infants' communicative behaviors

The first aim of the present study was to investigate if mothers of ELGA infants differed from mothers of FT infants in the contingency and relevancy of the communicative responses provided to their infant's gestures and vocal utterances.

Our findings showed that ELGA infants demonstrated significantly less advanced communicative skills, relative to FT infants, at 12 months. This study highlighted in ELGA infants, with respect to FT infants, a lower frequency of pointing/giving gestures, whereas a higher frequency of requesting/showing gestures, which are deictic gestures developmentally less advanced than pointing/giving, providing evidence for delayed trajectories of communication development in these infants. The above findings support the hypothesis that ELGA infants at 12 months have just started to use deictic gestures that particularly involve the social partner (Blake, O'Rourke, & Borzellino, 1994; Caselli et al., 2012; Iverson et al., 1994; Sansavini, Bello et al., 2010) and representational gestures that document meaning construction and expression (Bates et al. 1979; Capirci & Volterra, 2008). Thus, ELGA infants appear to be delayed in the gestural communication with respect to typically developing infants. Regarding infant's vocal utterances, in line with other studies that did not find differences between preterm and FT infants at 12 months in vocalization and babbling (Stolt et al., 2012) and in word production (D'Odorico et al., 2011; Sansavini, Guarini, Savini, et al., 2011), we did not find differences between ELGA and FT infants at 12 months. An explanation for this finding might be that the longer environmental auditory experience characterizing ELGA infants, born about three months in advance with respect to FT

infants, support their vocal development (Oller et al., 1994). Although it is possible that vocal production delays might emerge at subsequent ages, gestures appear a more important index of communicative delay in ELGA infants than vocal utterances, at 12 months. These findings bring evidence of the importance of analyzing gestures as main communication predictive indexes at 12 months (Bavin et al., 2008; Iverson & Goldin-Meadow, 2005; Reilly et al., 2009; Sansavini, Guarini, Savini, et al., 2011).

Despite the less advanced communicative abilities found in the ELGA infants, the mothers of these infants did not differ from the mothers of the FT infants in the contingency and relevancy of the responses provided to the infant's communicative behaviors at 12 months. With regard to contingency of the maternal responses, results indicated that the mothers of ELGA infants demonstrated prompt responses when interacting with their infants, similarly to the mothers of FT infants. Therefore, despite the communication problems of the ELGA infants and the resulting difficulties that the mothers of ELGA infants could meet for understanding their infants' communicative signals, these mothers were attentive to provide timely responses to their infants, as the mothers of FT infants did.

In addition, the mothers of the ELGA infants, as the mothers of the FT infants, provided a higher percentage of relevant responses than non-relevant responses, revealing that their responses to infant's communicative behaviors assume a supportive role in fostering language development. The maternal responses of the two groups were also comparable for grade of relevancy: in both groups, the most frequent maternal relevant responses were with no label, followed by responses with one label and with more labels. Thus, in both groups, relevant responses characterized by low grade of relevancy were predominant with respect to labeling responses. The prevalence of relevant responses with no label at 12 months of age could depend on the infant's gestural and vocal behaviors whose referents may not be easily identified by mothers at this age, both in ELGA and FT infants. It is possible that the maternal labeling of infant's gestures and vocal utterances, and thus

the grade of relevancy of the maternal responses, increases during the subsequent months, when the infant's communicative behaviors become more advanced and intelligible (Goldin-Meadow et al., 2007; Gros-Louis et al., 2006; Kishimoto et al., 2007).

In light of these results, our first hypothesis that less advanced communicative behaviors of ELGA infants could reduce the frequency of mother-ELGA infant effective exchanges and the opportunities, for these mothers, to provide relevant responses to their infants was not confirmed. These findings are in line with some (Barratt et al., 1992; Landry et al., 1997; Rahkonen et al., 2014), but not all (Reissland & Stephenson, 1999; Salerni et al., 2007), previous studies that, while examining several maternal communicative behaviors in mothers of preterm infants, did not find differences between these mothers and mothers of FT infants. As reported by Korja and colleagues (2012), studies indicate that during the first six months after birth maternal behavior during interaction seems to be at risk; however, differences in some maternal variables (e.g. facial responsiveness, smiling, controlling style) clearly decrease after the infant reaches a corrected age of 12 months. Our data add new evidence in this direction suggesting that extremely preterm birth does not negatively interfere with contingency and relevancy of the maternal communicative responses provided to the infant's communicative behaviors, at least at 12 months of corrected age.

Our opposite hypothesis appears instead to be confirmed. Despite the less advanced communicative behaviors of ELGA infants, their mothers seem to be particularly vigilant about scaffolding language. The mothers of ELGA infants might be concerned about their infants' risk for communicative delays and, thus, may pay particular attention to infant's gestures and vocal utterances, providing relevant responses. Two factors may have contributed to this result. First, the reduction of maternal stress and the increase of maternal self-esteem, as a result of interventions during neonatal hospitalization and transition to home (Meyer, Garcia Coll, Lester, Zachariah Boukydis, McDonough, & Oh, 1994; Trombini et al., 2008), may have helped to increase the positive maternal interactive behaviors. Second, the early counseling, provided to the parents of

ELGA infants during the medical and psychological follow-up program, realized at the Unit of Neonatology by a trained neuropsychological and neonatological team, for learning effective communicative strategies, may have improved the mothers' competences in reading the infants' communicative behaviors and in responding appropriately to them.

5.4.2 Reciprocal influences between maternal and infant communicative behaviors

The second aim of the present study was to shed light on the relationships between ELGA infant's communication and maternal responses in a view of development that acknowledges the bidirectional nature of parent-infant interaction (Sameroff, 2009; Leezenbaum et al., 2014). Consistent with this view, we highlighted interesting relationships between maternal relevant responses and infant's communication skills at 12 months of age, in particular in the mother-ELGA infant dyads. There were three main findings. The first concerned the relationship that we found between maternal contingent relevant responses and infant's spontaneous gestures. Specifically, we highlighted positive and strict relationships between relevant responses with more labels and both the pointing/giving gestures in the ELGA dyads, and the conventional gestures in the FT dyads. In line with the studies in typical development, showing caregivers more likely to translate the infant's more developmentally advanced gestures with respect to less advanced gestures (Kishimoto et al., 2007; Masur, 1982; Olson & Masur, 2011), these findings show, for the first time, the same tendency also in mothers of ELGA infants. The mothers of both groups, appear more likely to provide relevant responses with more labels to the infant's advanced gestures (i.e. pointing/giving in ELGA infants, conventional gestures in FT infants) relative to the most frequent but less advanced gestures of their infants (i.e. requesting/showing in both groups). Thus, by virtue of influences that the infant's communicative behaviors may have on the input of their caregiver, it is possible to suppose that a delay in the gestural development exhibited by ELGA infants (i.e. a low production of pointing/giving gestures at 12 months of corrected age, as emerged in this study) may reduce the

opportunities for the adults to provide translations of their infants' gestures, with cascading effects on later linguistic development (Goldin-Meadow et al., 2007; Olson & Masur, 2015).

Second, a similar maternal pattern was observed toward the infant's vocal productions. The positive relationship in ELGA dyads between maternal relevant responses with more labels and infant's spontaneous words at 12 months shows the tendency of the mothers of ELGA infants to translate the more advanced vocal utterances of their infants (i.e. words relative to nonword vocal utterances). Moreover, interestingly, the maternal responses with no label in ELGA dyads were negatively related to infant's spontaneous words. These findings could mean that the mothers of ELGA infants, when their infants produce first words, calibrate their responses reducing responses with no label and increasing repeated labeling responses (e.g. mothers rephrase correctly and many times the infant's word). Thus, despite the infant's first words are not yet so intelligible at 12 months, these mothers appear pretty good at guessing their infant's meaning. These relationships could also mean that the more the mothers of ELGA infants use responses with more labels, the more their infants produce words at 12 months of corrected age. At our knowledge, this is the first study that has investigated the associations between infant's communicative development and maternal responses distinguishing those with one label from those with more labels. Although further research is needed in order to generalize our findings, we can suppose that maternal responses characterized by repeated labeling may have important influences on infant word production.

In line with the expectations, the third interesting finding concerned the relationships that we found, specifically in ELGA dyads, between maternal labeling responses and infant's language skills evaluated with BSID-III at 12 months. Specifically, the maternal relevant responses with only one label were related to the ELGA infant's expressive communication, while the maternal relevant responses with more labels were associated with the ELGA infant's receptive skills. Therefore, a stricter association between maternal responses and infant's communicative behaviors was found in

ELGA dyads than in FT dyads. We can suppose that maternal concern about infant's language learning may make ELGA mothers very attentive to adapt their responses to their infants' receptive and expressive communication levels. A particularly novel and interesting finding from this study has to do with the maternal responses with more labels that appear strictly related to the ELGA infant's language comprehension, at 12 months. This relationship may mean that not only the spontaneous communicative abilities but even the comprehension skills of the ELGA infants can shape the language input they receive. Consistent with evidence in typically developing population (Olson & Masur, 2015; Tomasello et al., 2007), all these relationships seem to suggest that the maternal labeling responses could have important influences on the infant language development, particularly in infants at risk for communication delay.

All these findings bring evidence in favor of our hypothesis that associations among maternal relevant responses and infant's communicative-linguistic skills may be more evident in the ELGA dyads relative to the FT dyads. As some authors suggested (Landry et al., 1997), it is possible that maternal contingent and relevant communicative behaviors may be particularly important across the first year of life for ELGA infants, because of their early communication difficulties.

The second year of life is a critical period for language acquisition because an increase in the receptive and expressive vocabulary is usually observed in typical development (Bello, Giannantoni, Pettenati, Stefanini, & Caselli, 2012; Caselli et al., 2012; Gershkoff-Stowe & Hahn, 2013). Moreover, low expressive vocabulary at 24 months has often been used as an indicator of delayed language onset and for early identifying children as late talkers (Dale, Price, Bishop, & Plomin, 2003; Girolametto, Pearce, & Weitzman, 1996). The important contribution of this study is the examination of the receptive and expressive language skills exhibited at 24 months by the ELGA infants, relative to the FT peers. Differences between the two groups were identified in the composite language score, with the ELGA infants performing more poorly than the FT infants. Since we found less advanced language skills in ELGA infants, relative to FT infants, at 24 but not

at 12 months, we can suppose a slower development of communicative abilities in the ELGA infants, compared with the FT peers (Sansavini, Guarini, Savini, et al., 2011), during the second year with differences in language functions between ELGA and FT infants becoming more evident from the second year of life (Fasolo, D'Odorico, Costantini, & Cassibba, 2010). A potentially consequence of reduced production of pointing/giving and representational gestures observed among the ELGA infants in this study at 12 months is that mothers of ELGA infants may have fewer opportunities to provide labeling responses. Thus, this hypothetical scenario may explain the less advanced language abilities emerged in ELGA infants at 24 months. Viewed from this perspective, and as other authors suggest in infants at high risk of autism (Leezenbaum et al., 2014), the less advanced gestural development exhibited by the ELGA infants may have cascading effects on later language development by virtue of alterations in the input that infants may elicit from their caregivers. However, it is also possible that the BSID-III language scale is able to detect infants with less advanced language development from 24 months of age and not early when gestures more than words are a more relevant index of communication development; differently from the observation of the mother-infant interaction, which appears able to identify differences between the two groups as early as 12 months, standardized assessment tools could show limitations for identification of early developmental delays (Lobo et al., 2013).

Interestingly for purposes of the present study is the relationship that we found between maternal relevant responses with more labels at 12 months and infant's expressive language at 24 months, specifically in ELGA dyads. As it is known in typical developing population, by responding with labeling, the mother provides the object's label while the infant's attention is actively focused on its referent; moments of this sort are optimal for word learning (Goldin-Meadow et al., 2007; Olson & Masur, 2015; Tomasello et al., 2007). Therefore, the relationship between maternal repeated labeling responses and infants' expressive language, emerged specifically in ELGA dyads, represented a particularly interesting finding. This relationship

suggested that the maternal strategy of responding with more labels to the infant's early communicative behaviors could support the infant's expressive language development, more than maternal relevant responses without label or with only one label, in the ELGA dyads. The regression analysis confirmed this hypothesis, as we will discuss in the next paragraph.

Less advanced development in ELGA infants, relative to FT infants, was also observed in cognitive skills assessed with the BSID-III, both at 12 and 24 months of age. These findings suggest that in the first two years of life, ELGA infants show apparent difficulties, relative to FT infants, in cognitive performances. However, no significant associations between maternal contingent relevant responses and cognitive skills were found for either group at both ages. The maternal responses investigated in this work were strictly related to infant's communication behaviors rather than other infants' skills, such as cognitive skills, not observed in this study (e.g. infant's actions with objects not combined with communicative behaviors). Thus, we cannot exclude that others measures of maternal responsiveness could be related with neurocognitive outcomes in this clinical population at following ages (Rahkonen et al., 2014).

5.4.3 Role of the maternal repeated labeling on the infant's expressive language outcomes

The third aim of the present study was to analyze whether the early maternal repeated labeling predicted the infant's expressive language at 24 months. There is recent evidence that infant's communicative behaviors are related to their vocabulary outcomes mainly because they elicit responses that facilitate vocabulary acquisition (Olson & Masur, 2015), and that the maternal labeling is an important predictor of infant language development (Carpenter, Nagell, & Tomasello, 1998; Olson & Masur, 2015). In line with these findings in typically developing infants, the present study, for the first time in a mothers-ELGA infants group, proves that maternal repeated labeling to infant's communicative behaviors play a causal role in expressive language learning of ELGA infants. Interestingly, considering together maternal repeated labeling responses, neonatal status (i.e.

being ELGA versus FT) and infants' expressive communication skills at 12 months, only the proportion of repeated labeling responses that the mothers produced following infants' communicative behaviors was a significant independent predictor of infants' expressive language at 24 months.

Our findings bring thus new evidence about the positive effect of maternal contingent and relevant responses on later language development, since previous longitudinal studies run on preterm populations have mainly investigated associations among maternal responsive behaviors and infant competencies at subsequent ages (Bozzette, 2007; Landry et al., 1997; Rahkonen et al., 2014) and have not examined maternal contingent and relevant responses as a predictive factor.

Importantly, the role of the maternal repeated labeling on language development appears more important than individual factors, such as the expressive communication level at the first year of age and being born at term. Thus, these results provide an important contribution for identifying early factors that could influence the language outcomes of this clinical population. It is possible to suppose that environmental factors, such as the relevant linguistic feedback that the infants receive from their surroundings, may be particularly important in populations at risk of language delays, as ELGA infants, for supporting their language development (Landry et al., 1997).

The implications of these findings for developmental research and clinical applications will be considered below.

6. GENERAL DISCUSSION

The primary objective of this work was to examine the early communicative development in ELGA infants, researching potential profile of risk that could be used to the early identification of language delays and impairments in these infants. This is a relevant contribution in the current research landscape, given that the extremely preterm infants are the preterm infants at highest risk for language delays (Sansavini, Guarini, & Caselli, 2011) and rarely researchers have investigated, in the first years of life, early markers of subsequent language impairment in this clinical population. This work was guided by the conceptual framework of DST (Thelen & Smith, 1994) which asserts that complex phenomena development (e.g., language system), as well as their atypical development, cannot be fully understood by dissecting the system into its constituent parts and by excluding its interaction with the broader social environment. In fact, the individual components are embedded within the fabric of the whole system and are built upon a continual interplay with the social context (Iverson, 2010). Thus, the relationships among intra-individual components and between these and environmental factors were of particular interest in this research.

Specifically, Study 1 was designed to evaluate multimodal communication abilities, i.e. gestures, vocal productions, and the capacity to coordinate gesture, speech and gaze toward the mother in ELGA infants relative to FT infants, with the aim to verify whether ELGA infants

differed by FT infants in these abilities as early as 12 months of age. Motivated from the recent surge of interest on links between action and language (Hill, 2001; Iverson, 2010; Leonard & Hill, 2014), investigating the nature of the early relationships between gestural abilities and motor skills in these two groups, was of particular interest as well.

Consistent with DST and with the point of view of some authors (Landry et al., 1997; Karmiloff-Smith, 2009), for infants at risk for developmental disabilities such as ELGA infants, variability in the course of communicative development may due to both individual characteristics and characteristics of their surroundings. As we have seen by reviewing the literature, to date, none has focused on ELGA populations integrating together infant's and maternal communicative behaviors. Thus, in Study 2 we examined how the mothers of ELGA infants responded to their infants' communicative signals, relative to mothers of FT infants. Of particular interest was the nature of the reciprocal influences between mother's and ELGA infant's communicative behaviors, and the predictive role of the maternal relevant responses on language development of these infants.

6.1 Characterizing the early mother-ELGA infant dyads communicative development

6.1.1 Early potential indexes of communicative difficulties in ELGA infants

As described in the introduction at the present work, the first two research questions that we addressed were the following. Do the young ELGA infants show less advanced multimodal communicative abilities, compared with FT infants, at the first year of age? Are the communicative and motor domains related in the ELGA group?

Study 1 revealed that giving and representational gestures were produced at a lower rate by ELGA infants, and that pointing gestures and words were produced by a lower percentage of ELGA infants, relative to FT infants, at 12 months. By contrast, the communicative coordinations (i.e. combinations among gestures, vocal utterances and gaze toward the mother) did not distinguish

ELGA infants from FT infants at this age. Less advanced gross and fine motor skills were also found in ELGA infants, relative to FT infants. Interestingly, the best gestural predictors of language development, i.e. pointing and representational gestures (Bavin et al., 2008; Capirci & Volterra, 2008; Caselli et al., 2012; Iverson & Goldin-Meadow, 2005; Reilly et al., 2009), were related to fine motor skills in the ELGA group.

The findings from the current investigation suggest thus that, among all communicative behaviors that the infants show at 12 months of age, gestures and first words appear to be the best potential indexes of communicative delays in ELGA infants. However, the lack of significant group differences in the frequency of communicative coordinations is an unexpected result: the ELGA infants seem to show a typical profile in this type of multimodal communication at 12 months. Nevertheless, we cannot exclude that less advanced capacities to coordinate gesture-speech-gaze toward the mother may emerge at subsequent ages, as highlighted in other atypically developing populations (Winder et al., 2013).

The relationships that we highlighted between gestural and fine motor domains, are an interesting contribution of the present research that could bring evidence, as suggested by recent theories (Capirci, Contaldo, Caselli, & Volterra, 2005; McNeill, 2005; Iverson & Thelen, 1999; Sansavini, Bello, et al., 2010), about the role of early motor and gestural productions on the construction of the representation of meaning. Furthermore, given that we found stricter relationships in the ELGA group, relative to the FT group, this research seems to bring also evidence that atypicality in one ability can have ramifications in others abilities in the ELGA population, in line with findings in atypical developing populations (Karmiloff-Smith, 2009; Sansavini, Guarini & Caselli, 2011). Thus, we can suppose that the delays in motor experiences that we found in the ELGA infants may constrain their language acquisitions (LeBarton & Iverson, 2013). These findings leave open some questions: for example, which specific fine motor difficulties (e.g., difficulties of eye-hand coordination, manipulation problems) influence the

communicative development of ELGA infants, or whether there are inter-domain relationships also between motor and communication receptive abilities. Although these findings are only an initial evidence of the associations between motor and communicative development in ELGA infants, they point out the importance of studying communicative development of this clinical population according to the DST perspective.

Regarding methodological aspects, this research provides a relevant contribution that could be important in informing clinical practice, namely, the assessment of ELGA infants. The direct observation of spontaneous communicative behaviors and the coding scheme employed in Study 1 for detecting infant's communicative behaviors appear a more eligible measure for identifying atypical profiles at 12 months of age, than parental report measures and structured tools (Lobo et al., 2013). Then, the further qualitative analysis of the infant's communicative behaviors that we conducted in the Study 2, in which less advanced communicative behaviors were merged and distinguished from more advanced communicative behaviors, helped to better define the delayed profile of ELGA infants. With regard to the assessment at subsequent ages (e.g., second and third year of life), we suppose that even more structured measures may be effective. Specifically, as highlighted in Study 2 and consistent with other authors (Perra, McGowan, Grunau, Doran, Craig, et al., 2015; Sansavini et al., 2014), the BSID-III appears an appropriate measure for identifying less advanced language abilities in ELGA infants, relative to FT infants, at 24 months of age.

In sum, this research provides a detailed characterization of the early communicative development of the preterm infants at highest risk for language delays, thanks to the homogeneous group of healthy ELGA infants involved and to the wide description of their vocal and gestural behaviors in relation to motor skills. Continuing to conduct studies in this way, namely focusing on ELGA infants and thus minimizing the variability of the bio-medical factors, we can reduce the possibility of conflicting data and better understand how the extremely preterm birth affects communication development. However, only when we integrate individual and environmental

components, as we have done in Study 2, we obtain a more complete picture of the factors involved in the communicative development of these infants.

6.1.2 Role of maternal communicative responses on language development of ELGA infants

After focusing on the infant, the next logical question that we addressed in this work was whether the communicative strategies of the mothers of ELGA infants could affect the language development of their infants.

As described in the introduction, a third and a fourth research question guided thus this work. Do the mothers of ELGA infants differ by the mothers of FT infants in the way in which they respond to their infants' communicative behaviors? Do the maternal responses influence the communicative-linguistic development of ELGA infants?

The answer to the third question is “no”. In part unexpectedly, Study 2 revealed no significant group differences for the contingency and relevancy of the responses that the mothers provided to their infants' communicative signals. Thus, these findings demonstrate that the extremely preterm birth does not seem to influence these types of maternal variables. Despite the prevalence of requesting/showing gestures in their infants, namely gestures able to elicit fewer maternal relevant responses than pointing/giving gestures (Kishimoto et al., 2007; Olson & Masur, 2011), the mothers of ELGA infants are as responsive as the mothers of FT infants. In other words, the less advanced communicative abilities of ELGA infants do not seem to reduce the opportunities for these mothers to provide prompt and relevant responses useful for language development. However, we cannot exclude that differences between the two groups of mothers could emerge at subsequent ages. As suggested by some authors (Sansavini, Guarini, Savini, et al., 2011) and as our findings seem to confirm, the language difficulties of ELGA infants appear to increase over time. Thus, it is possible that an increasing gap between the mothers of the two groups could later emerge as well. Moreover, although the maternal variables investigated in this research are useful in initial evaluations, it

would be interesting to run an additional analysis aimed to investigate for what types of infant's communicative behaviors the mothers of ELGA infants more frequently respond. As highlighted by some studies both in typically and atypically developing populations (Gros-Louis et al., 2006; Leezenbaum et al., 2014; Olson & Masur, 2011), the mothers are more likely to respond with rich communicative input (i.e., translations and labeling responses) to infant's behaviors that are more developmentally advanced. Thus, to distinguish between maternal responses specifically directed to the infant's more advanced communicative behaviors (e.g., pointing/giving, words) and those directed to the less advanced communicative behaviors (e.g., requesting/showing, vocalizations) could provide additional information about the maternal communicative strategies of the two groups.

An interesting result of the present research concerns the associations between the infant's communicative behaviors and the maternal relevant responses that we found in the ELGA dyads. These relationships suggest that the mothers of the ELGA infants are particularly sensitive to the developmental level and quality of their infants' communicative behaviors, providing meaningful responses despite the little advanced communicative signals of their infants. We considered some possible explanations of this result. Some authors have described the positive effects of the parent-implemented intervention on language development of late-talking children (Girolametto et al., 1996). Thus, it is possible that the frequent clinical follow-up visits, offered to these families since infant's birth by the Day-Hospital of the Unit of Neonatology at Bologna University, helped to obtain this result. During these follow-up visits, clinicians encouraged caregivers to pay attention to all communicative signals of their infants and to respond them in an appropriate manner. If so, the lack of significant group differences appears very interesting and particularly encouraging. However, these considerations remain merely hypothetical reflections. In fact, an evidence-based investigation of the positive effects of these follow-up visits on the maternal communicative responses of these mothers would be needed to confirm this hypothesis. Also controlling for other

maternal variables, such as maternal stress and anxiety, could add useful information to better explain the lack of differences between these two groups of mothers (Coletti, Caravale, Gasparini, Franco, Campi, & Dotta, 2015).

Finally, a novel and important contribution of the present research was to consider the role of the maternal relevant responses on the ELGA and FT infants' language development. As widely described in the introduction of the present work, extensive research on typically developing populations, has shown that mothers who respond to the infant's focus of attention and communicative behaviors with relevant responses can promote infant's language acquisition (Goldin-Meadow et al., 2007; Tamis-LeMonda et al., 2001; Tomasello et al., 2007). This is the first research that has investigated the effects of maternal relevant responses on language development in mother-ELGA infant dyads. Interestingly, we can answer “yes” to our fourth research question. In fact, Study 2 found that the maternal responses characterized by more labels at 12 months predicted the infants' expressive language at 24 months in the ELGA dyads. Thus, the expressive communication outcomes of the ELGA infants appear depending on the quality of the maternal responses rather than on the group membership and on infant's expressive skills at 12 months. Certainly, the extent to which researchers can understand the characteristics of early communicative abilities in ELGA infants and how these abilities are affected by environmental inputs will have important implications for how treatment programs are designed and implemented.

6.2 Limitations and future directions

Limitations of this work should be noted. First, data from Study 1 are limited to a single age point. There is a need for longitudinal studies aimed at understanding the extent to which gesture and word production abilities at 12 months contribute to the prediction of language outcomes and provide useful information regarding their value as indices of risk for future language delays and

impairment in the ELGA population. Moreover, longitudinal studies with several points of assessment permit the identification of differences in the developmental trajectories of ELGA and FT infants (Sansavini et al., 2014; Thomas, Annaz, Ansari, Scerif, Jarrold, & Karmiloff-Smith, 2009). As noted above, it is possible that group differences in communicative coordinations were not detected due to the young age of the infants in the study. Thus, subsequent points of assessment are needed to fully understand the development of this types of behaviors in ELGA infants. The present research, being part of a larger longitudinal study, is a step in this direction.

Second, data from Study 2 regarding the maternal communicative responses are limited to a single point of observation as well. Future research is necessary to verify whether mothers of ELGA infants maintain or change over time this pattern of responses. For example, at subsequent ages we could observe group changes in pattern of maternal responses that may reflect group differences in the ELGA and FT infants' communicative quality. Moreover, Study 2 did not examine mothers' overall communication directed toward their infants. It is possible that mothers of ELGA and FT infants provide different types of interactive behaviors that are not necessarily in response to their infants' communicative behaviors, as shown in a recent study demonstrating that mother-ELGA infant dyads at 12 months experience less frequent symmetrical and more frequent unilateral co-regulation patterns and less positive and more neutral affective intensity with respect to mother-FT infant dyads (Sansavini, Zavagli, Guarini, Savini, Alessandroni, & Faldella, 2015).

Third, although this research has contributed to our understanding of the relationship among gestures and motor skills in ELGA infants, it did not include a description of the specific types of fine motor behaviors characterizing the two groups. This could be achieved in future research by conducting studies assessing the relative contributions of different types of fine motor skills (e.g., types of object grasping and manipulation, functional hand skills, and eye-hand coordination) to communicative-linguistic development in ELGA infants, by using, besides broad developmental assessment tools, specific motor assessments such as the Alberta Infant Motor Scales (Piper &

Darrah, 1994), observational motor coding schemes, and experimental techniques employing sensor-based technology. Moreover, more longitudinal research is needed to elucidate the way in which early motor delays contribute to language and communication impairment in ELGA infants.

Fourth, the sample size utilized in the present research was small, since it was focused on mother-ELGA infant dyads. The generalization of our findings should be carefully considered. The limited sample size may have impacted the ability to detect some differences in communicative behaviors between ELGA and FT infants and in communicative responses between the two groups of mothers. Replication of the present findings with larger samples is clearly needed in the future.

While more longitudinal research with larger sample sizes, further analyses of behavior, and additional clinical comparison groups, for example with VLGA infants, is needed to generalize the findings documented here, these results have clear clinical implications that will be considered below.

6.3 Clinical implications

6.3.1 Integrating infant's spontaneous communicative abilities, motor skills and maternal communicative responses in the early assessment of mother-ELGA infant dyads

Findings from this research suggest that the assessment of the ELGA infant development should be particularly intensive and must incorporate both several neuropsychological domains in relation to each other and maternal communicative modalities. Specifically, clinical assessment of ELGA infants must address spontaneous communication abilities, in particular gestures and word production, as early as the first year of life. Instead, the potential usefulness of communicative coordinations, in identifying ELGA infants at risk for communicative delays, might emerge at subsequent ages. Our findings also show the value of examining early development in the motor domain. In infants with communicative difficulties, delays in motor development may be

conceptualized as indexes of general delayed maturation, whereas they should not be undervalued in early assessments (Iverson & Braddock, 2011). In particular, fine motor delays should be carefully considered, for their potential associations with communicative delays. In fact, fine motor difficulties are evident in a substantial proportion of ELGA infants, and they can have significant negative consequences for infants' gestural abilities and, thus, for the early communicative exchanges with the caregiver. Importantly, screening projects should be conducted to identify infants performing below average on motor development, follow them longitudinally, and thus prevent risks for later neurodevelopmental disorders (Leonard & Hill, 2014). The current investigation also points to the importance of the examination of the maternal relevant responses to fully interpret the language outcomes of ELGA infants. The direct observation of the maternal communicative responses should be therefore included in the standard assessment of the mother-ELGA infant dyads. It may help to understand, for example, why some ELGA infants recuperate the linguistic divergence with respect to FT infants while other ELGA infants do not.

Our findings indicate the importance of considering all these infants' and maternal communicative behaviors as they occur naturally in a familiar setting. In particular, observation of the ELGA infant's spontaneous communication should be incorporated into clinical assessment and practice because it seems very effective in detecting the quality of the early communicative abilities.

In light of the above, it appears urgent to develop measures that can be rated by clinical staff during follow-up visits, able to capture information about infant's and mother's spontaneous communicative behaviors. To date, it is not yet practical to code during medical office visits. Future research is necessary to adapt coding schemes, as those employed in this work, to clinical assessment. The main challenge is to find a method that allows an “on-line” coding (i.e. live coding) and/or which is compatible with the clinical tight deadlines.

6.3.2 Early interventions focused on mother-ELGA infant dyads could prevent language delays

The accurate identification of less advanced communicative and motor abilities in the first year of life is an ongoing challenge, for ensuring the provision of effective early intervention services to ELGA infants and their families (Lobo & Galloway, 2013).

It is understood that there are several practical issues that may hinder the early taking in charge of the ELGA infants, including time, money, and clinical resources of the health services. Nevertheless, it is known that the positive effects of early interventions may be enhanced by the high degree of cerebral plasticity of the infant brain, that is the capacity to respond in a dynamic manner to the environment and experience through the modification of neural circuitry (Anderson, Spencer-Smith, & Wood, 2011). In addition, it is evident the lifetime cost for society of caring for a preterm child. For example, Chaikind and Corman (1991), focusing on LBW children, investigated the impact of that condition on special education costs in the United States. Using a sample of approximately 8000 children aged 6-15 years, they calculated the probability of a child attending special education. LBW children were almost 50% more likely to require special education than children who were of normal birthweight. Therefore, we can suppose that early interventions in ELGA infants, not only could have positive effects on infants' later gains and on families' emotional well-being, but also might reduce the long-term costs for society (Chaikind & Corman, 1991).

In the light of our findings, it might be very useful to develop an intervention approach designed to enhance the gestural and words productions of ELGA infants. Also early interventions on motor skills might bring benefit, not only on these skills which appear compromised in the first year of life, but also on communicative development. For example, postural control, that is the ability to control the body's position in space for stability and orientation, is not only important for development of gross and fine motor skills, but it also appears effective for cognitive development (Spittle et al., 2009). To date, there is little evidence about the effectiveness of early communicative and motor interventions on the improvement of the linguistic abilities of ELGA infants. Thus, more

evidence-based research is needed to verify the long-term effects of early interventions as those mentioned.

In addition to infant-centered intervention programs, our results also emphasize the importance of specific programs designed for the mothers of these infants. Our results seem to suggest that the more the mothers of ELGA infants provide repeated labeling responses to their infants' communicative behaviors, the more their infants develop expressive language abilities. Thus, we think that clinical follow-up visits for ELGA infants should also include counseling for their mothers, aimed to increase their awareness about the importance to provide repeated labeling responses to their infants. Moreover, consistent with growing evidence that found that early interventions involving parents can improve neurodevelopmental outcomes of preterm infants (Spittle et al., 2009; for review, see Vanderveen, Bassler, Robertson, & Kirpalani, 2009), we presume that early parent-implemented interventions that encourage mothers of ELGA infants to provide relevant responses to their infants' communicative behaviors could support the language development. Although the need for evidence-based research to verify whether these types of interventions can promote recovery processes in ELGA infants is critical, our findings are suggestive. In fact, they point out the importance of interventions that include naturalistic interaction with familiar caregivers and, for the first time, they put the maternal repeated labeling responses among the potential intervention strategies for supporting the language development of ELGA infants.

7. GENERAL CONCLUSION

Differently by the vast majority of the studies investigating the communicative behaviors of heterogeneous preterm groups, the present research focused on a homogeneous group of healthy ELGA infants and their mothers. In this way, we could better understand the early effects of the extremely preterm birth on the communicative-linguistic development. Very rarely the communicative development of this clinical population has been studied as multimodal process, that is characterized by the dynamic interplay among individual components in interaction with the context, as the DST suggests (Thelen & Smith, 1994). Accordingly, understanding the phenomenon of atypical or delayed communication requires investigations in multiple modalities, the use of multiple methods and contexts, and assessments of interactions between individual and environmental components as they unfold over time (Iarocci & McDonald, 2006).

In the present research, incorporating measures derived from spontaneous mother-infant interactions, parent reports and structured evaluation, we could obtain a complete picture of the early multimodal communicative abilities of ELGA infants, highlighting early differences with respect to FT infants. In fact, the findings from the current investigation suggest that these ELGA infants demonstrate difficulties in the early communication, in particular in gestural modalities which appear in strict relationship with the fine motor skills, the latter also delayed.

These data also emphasize the importance of understanding ELGA infant development in the

context of social interaction. This research, for the first time, has considered together the infant's communicative behaviors and the maternal contingent and relevant responses to these behaviors. Our findings suggest that the mothers of ELGA infants do not appear at risk for providing less contingent and relevant responses relative to the mothers of FT infants. Despite the less advanced communicative behaviors of the ELGA infants could negatively influence the communicative exchanges with the mother, the mothers of these infants seem to took every opportunity to respond in appropriate manner to their infants' communicative behaviors. As reported by Korja and colleagues (2008), extremely preterm birth does not always negatively affect the quality of maternal behaviors, especially when continuous parenting support is available in the neonatal intensive care unit and after discharge from the hospital. Overall, the present findings highlight the predictive value of the maternal repeated labeling responses on the infant's language acquisitions. The impact of the repeated labeling underscores the combined influence of early infant and maternal factors in understanding ongoing language development in this clinical population. Interestingly, these findings suggest that the maternal repeated labeling may be helpful to reduce the communicative-linguistic delays of ELGA infants. As described in the present research, however, the nature of mother-ELGA infant interactions is reciprocal, whereby the contributions of both communicative partners are likely to influence and be influenced by the other at different ages of development. Thus, the causal pathway demonstrated here requires further investigation.

We therefore argued the importance, for the clinical practice, of monitoring gestures, words production and motor skills in ELGA infants, for identifying infants at risk for language delays and for implementation of specific interventions. The intervention programs should involve the family as valuable resource (Spittle et al., 2009), encouraging caregivers to provide appropriate responses, such as repeated labeling. Future evidence-based research is needed to understand what kind of intervention could help these infants to establish a solid foundation for later language gains.

In conclusion, we hope that these findings could be a starting point for identifying early

markers of language delays in ELGA infants and, subsequently, early targets for intervention within the context of parent–infant interaction.

8. TABLES

Table 1

Perinatal and socio-demographic characteristics of the ELGA and FT groups.

			ELGA	FT
			<i>n</i> = 20	<i>n</i> = 20
Infant's perinatal characteristics				
Gestational age (weeks)		<i>M (SD) range</i>	25.7 (1.4) 23-28	39.5 (1.2) 38-42
Birthweight (grams)		<i>M (SD) range</i>	803 (191) 509-1093	3476 (464) 2500-4200
Small for gestational age (SGA)		<i>N (%)</i>	2 (10)	-
Respiratory distress syndrome needing mechanical ventilation (RDS-MV)		<i>N (%)</i>	20 (100)	-
Hyperbilirubinemia treated with phototherapy		<i>N (%)</i>	14 (70)	-
Bronchopulmonary dysplasia (BPD)*		<i>N (%)</i>	13 (65)	-
Retinopathy of prematurity (ROP) of grade I or II		<i>N (%)</i>	13 (65)	-
Intra-ventricular hemorrhage (IVH) of grade I or II		<i>N (%)</i>	1 (5)	-
Hyperechogenicity (HE) of white matter^		<i>N (%)</i>	17 (85)	-
Socio-demographic characteristics				
Infant's gender	Male	<i>N (%)</i>	9 (45)	11 (55)
	Female	<i>N (%)</i>	11 (55)	9 (45)
Birth order	First born	<i>N (%)</i>	15 (75)	18 (90)
	Later born	<i>N (%)</i>	5 (25)	2 (10)
Maternal age (years)		<i>M (SD) range</i>	36.2 (4.8) 27-44	34.6 (3.1) 30-41
Maternal education	Basic/High school	<i>N (%)</i>	12 (60)	8 (40)
	University/Postgraduate	<i>N (%)</i>	8 (40)	12 (60)

*BPD was defined as need of supplemental oxygen at 36 weeks of postconceptional age.

^Persistent hyperechogenicity (HE) of white matter (≥ 14 days), but this had completely resolved by 3 months.

Table 2

Comparisons between ELGA and FT groups of the rate per 10 min of gestures (Mann-Whitney test) and of the number of infants producing gestures (Chi-Square test) at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> =20)	FT (<i>n</i> =20)	Mann-Whitney		
		<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>U</i>	<i>p</i>	<i>r</i>
Gestures	Requesting/Reaching	6.23 (6.79)	2.50 (1.90)	149	.167	.22
	Pointing	2.12 (3.98)	2.97 (3.74)	133.5	.070	.29
	Showing	1.54 (1.79)	1.38 (2.15)	173.5	.464	.12
	Giving	.26 (.70)	1.52 (2.44)	104	.004	.46
	Conventional	.52 (1.10)	1.12 (1.64)	162	.272	.17
	Representational	.28 (.69)	1.59 (2.41)	117.5	.015	.39
		<i>n</i> . (%)	<i>n</i> . (%)	Chi- Square		
				χ^2	<i>p</i>	<i>phi</i>
Gestures	Requesting/Reaching	18 (90%)	18 (90%)	-	1.000 [^]	-
	Pointing	12 (60%)	18 (90%)	4.80	.028	.35
	Showing	14 (70%)	12 (60%)	.44	.507	.11
	Giving	4 (20%)	13 (65%)	8.29	.004	.46
	Conventional	9 (45%)	11 (55%)	.40	.527	.10
	Representational	6 (30%)	12 (60%)	3.64	.057	.30

Significant results are in bold.

[^] Fisher exact test.

Table 3

Comparisons between ELGA and FT groups of the rate per 10 min of vocal utterances (Mann-Whitney test) and of the number of infants producing vocal utterances (Chi-Square test) at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> =20)	FT (<i>n</i> =20)	Mann-Whitney		
		<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>U</i>	<i>p</i>	<i>r</i>
Vocal utterances	Vocalization	12.58 (8.10)	14.72 (11.61)	185	.685	.01
	Babbling	4.52 (4.61)	6.66 (6.64)	155	.223	.19
	Word	.99 (2.74)	1.10 (2.12)	138	.076	.28
		<i>n.</i> (%)	<i>n.</i> (%)	Chi- Square		
				χ^2	<i>p</i>	<i>phi</i>
Vocal utterances	Vocalization	20 (100%)	19 (95%)	-	1.000 [^]	-
	Babbling	17 (85%)	20 (100%)	-	.231 [^]	-
	Word	7 (35%)	14 (70%)	4.91	.027	.35

Significant results are in bold.

[^] Fisher exact test.

Table 4

Comparisons between ELGA and FT groups of the rate per 10 min of communicative coordinations (Mann-Whitney test) and of the number of infants producing communicative coordinations (Chi-Square test) at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> =20)	FT (<i>n</i> =20)	Mann-Whitney		
		<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>U</i>	<i>p</i>	<i>r</i>
Communicative Coordinations	Gesture+gaze	3.02 (2.30)	2.51 (3.04)	153.5	.208	.20
	Vocalization/babbling + gaze and/or gesture	5.19 (4.08)	6.28 (5.92)	194.5	.882	.02
	Word + gaze and/or gesture	.38 (1.51)	.60 (1.22)	148	.074	.28
		<i>n</i> . (%)	<i>n</i> . (%)	Chi- Square		
				χ^2	<i>p</i>	<i>phi</i>
Communicative Coordinations	Gesture+gaze	19 (95%)	16 (80%)	-	.342 [^]	-
	Vocalization/babbling + gaze and/or gesture	20 (100%)	19 (95%)	-	1.000 [^]	-
	Word + gaze and/or gesture	3 (15%)	8 (40%)	3.14	.077	.28

Significant results are in bold.

[^] Fisher exact test.

Table 5

Comparisons between ELGA and FT groups on the BSID-III motor, cognitive and language composite and scaled scores with the ANOVA at 12 months (corrected age for ELGA infants).

	<i>ELGA (n = 20)</i>	<i>FT (n = 20)</i>	ANOVA		
	<i>M (SD)</i>	<i>M (SD)</i>	<i>F</i>	<i>p</i>	<i>ηp²</i>
Motor composite score	87.95 (13.24)	100.30 (11.47)	9.94	.003	.207
Fine motor scaled score	9.15 (1.81)	11.40 (2.19)	12.54	.001	.248
Gross motor scaled score	6.80 (3.46)	8.75 (2.49)	4.19	.048	.099
Cognitive composite score	94.50 (11.46)	104.75 (10.70)	8.55	.006	.184
Language composite score	97.10 (12.76)	103.55 (12.32)	2.64	.112	.065
Receptive scaled score	9.35 (2.66)	10.85 (2.92)	2.88	.098	.070
Expressive scaled score	9.60 (2.48)	10.40 (2.14)	1.19	.281	.030

Significant results are in bold.

Table 6

Comparisons between ELGA and FT groups on the PVB-GW Words Understood, Words Produced, and Total Gestures/Actions scores (Mann-Whitney test) at 12 months (corrected age for ELGA infants).

	ELGA (<i>n</i> =20)		FT (<i>n</i> =19)		Mann-Whitney		
	<i>M</i> (<i>SD</i>)	<i>Range</i>	<i>M</i> (<i>SD</i>)	<i>Range</i>	<i>U</i>	<i>p</i>	<i>r</i>
Words Understood	75.15 (53.89)	0-170	118.16 (87.13)	19-297	133	.109	.25
Words Produced	3.05 (3.41)	0-11	2.84 (4.37)	0-14	165.5	.479	.11
Gestures/Actions	16.40 (8.42)	6-33	24.21 (9.57)	9-40	99.5	.011	.40

Significant results are in bold.

Table 7

Spearman's correlation coefficients between gestures and BDIS-III fine and gross motor scaled scores and cognitive composite score in ELGA and FT groups, at 12 months (corrected age for ELGA infants).

		<i>ELGA (n = 20)</i>									<i>FT (n = 20)</i>								
		Fine motor (scaled score)			Gross motor (scaled score)			Cognitive (composite score)			Fine motor (scaled score)			Gross motor (scaled score)			Cognitive (composite score)		
		<i>rs</i>	<i>p</i>	<i>rs</i> ²	<i>rs</i>	<i>p</i>	<i>rs</i> ²	<i>rs</i>	<i>p</i>	<i>rs</i> ²	<i>rs</i>	<i>p</i>	<i>rs</i> ²	<i>rs</i>	<i>p</i>	<i>rs</i> ²	<i>rs</i>	<i>p</i>	<i>rs</i> ²
Gestures	Requesting/Reaching	-.039	.871	.002	.093	.697	.009	-.134	.574	.018	-.045	.851	.002	-.114	.631	.013	-.328	.158	.108
	Pointing	.631	.003	.398	.398	.082	.158	.423	.063	.179	.064	.787	.004	-.200	.397	.040	.147	.535	.022
	Showing	-.034	.886	.001	.218	.356	.047	-.089	.709	.008	.089	.710	.008	-.089	.709	.008	-.167	.481	.028
	Giving	.362	.117	.131	.073	.758	.005	.015	.949	.000	.446	.049	.199	.359	.120	.129	.187	.431	.035
	Conventional	-.357	.122	.127	-.173	.446	.030	-.347	.134	.120	.290	.215	.084	-.473	.035	.224	.624	.003	.389
	Representational	.623	.003	.388	.234	.320	.055	.446	.049	.199	.090	.705	.008	-.149	.531	.022	.333	.151	.111

Significant results are in bold.

Table 8

Comparisons between ELGA and FT groups on the rates per 10 minutes of gestures and vocal utterances (Mann-Whitney test), at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> =20)	FT (<i>n</i> =20)	Mann-Whitney		
		<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>U</i>	<i>p</i>	<i>r</i>
Gestures at 12 months	Requesting/Showing	7.78 (6.44)	3.88 (3.12)	125	.042	.32
	Pointing/Giving	2.39 (4.05)	4.49 (3.89)	97	.005	.44
	Conventional	.52 (1.10)	1.12 (1.64)	162	.272	.17
	Representational	.28 (.69)	1.59 (2.41)	117.5	.015	.39
Vocal utterances at 12 months	Nonword vocal utterance	1.71 (.99)	2.14 (1.51)	175.5	.507	.10
	Word	.99 (2.74)	1.10 (2.12)	138	.076	.28

Significant results are in bold.

Table 9

Comparisons between mothers of ELGA and FT infants on the rates per 10 min and percentages of maternal responses to infant's communicative behaviors with the ANOVA, at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> =20)		FT (<i>n</i> =20)		ANOVA*		
		<i>Rates per 10 min</i>	<i>%</i>	<i>Rates per 10 min</i>	<i>%</i>	<i>F</i>	<i>p</i>	<i>ηp²</i>
<i>Maternal responses</i>		<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>			
Non-contingent responses		8.45 (6.17)	31 (14)	9.92 (7.24)	34 (12)	.523	.474	.014
Contingent non-relevant responses		2.54 (2.11)	11 (9)	2.31 (1.97)	8 (5)	.957	.334	.025
Contingent relevant responses	With no label	9.86 (5.00)	39 (11)	10.53 (6.54)	37 (10)	.580	.451	.015
	With one label	2.97 (1.88)	12 (5)	3.42 (2.01)	13 (9)	.549	.463	.014
	With more labels	1.96 (2.28)	7 (6)	2.07 (1.54)	8 (5)	.024	.878	.001

*ANOVA conducted on the percentages.

Table 10

Comparisons between ELGA and FT groups on the BSID-III language composite and scaled scores, cognitive composite scores and motor composite and scaled scores with the ANOVA, at 24 months (corrected age for ELGA infants).

	<i>ELGA (n = 20)</i>	<i>FT (n = 18)</i>	ANOVA		
	<i>M (SD)</i>	<i>M (SD)</i>	<i>F</i>	<i>p</i>	<i>ηp²</i>
Language composite score	93.65 (15.27)	102.67 (10.75)	4.34	.045	.107
Receptive scaled score	10.20 (3.25)	11.50 (2.81)	1.72	.198	.046
Expressive scaled score	8.10 (2.79)	9.33 (2.33)	2.16	.150	.057
Cognitive composite score	85.50 (7.24)	96.67 (8.57)	18.94	<.001	.345
Motor composite score	83.95 (7.17)	101.56 (10.14)	38.79	<.001	.519
Fine motor scaled score	8.30 (1.63)	11.33 (1.91)	27.97	<.001	.437
Gross motor scaled score	6.35 (1.09)	9.11 (1.97)	29.44	<.001	.450

Significant results are in bold.

Table 11

Spearman's correlation coefficients between the types of contingent salient maternal response at 12 months and infant's spontaneous gestures and vocal utterances in ELGA and FT dyads, at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> = 20)										FT (<i>n</i> = 20)													
		Gesture at 12 months				Vocal utterance at 12 months						Gesture at 12 months				Vocal utterance at 12 months									
		Requesting/ Showing		Pointing/ Giving		Conventional		Representational		Nonword		Word		Requesting/ Showing		Pointing/ Giving		Conventional		Representational		Nonword		Word	
		<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>	<i>rs</i>	<i>p</i>
Contingent relevant responses	With no label	.319	.171	-.417	.068	-.058	.809	-.150	.527	-.205	.387	-.515	.020	.135	.571	.147	.537	-.098	.682	-.060	.802	.163	.492	-.211	.371
	With one label	-.128	.591	.135	.570	-.309	.184	.375	.103	-.179	.450	.242	.305	-.159	.503	.133	.576	.327	.160	.182	.442	-.285	.224	-.246	.295
	With more labels	-.229	.332	.503	.024	.226	.337	.119	.617	-.141	.552	.475	.034	.124	.603	.198	.403	.554	.011	.108	.650	-.143	.548	.039	.871

Significant results are in bold.

Table 12

Pearson's correlation coefficients between the types of salient maternal response and BDIS-III receptive and expressive language scaled scores and cognitive composite score in ELGA and FT dyads, at 12 months (corrected age for ELGA infants).

		ELGA (<i>n</i> = 20)						FT (<i>n</i> = 20)					
		Receptive language (scaled score) at 12 months		Expressive language (scaled score) at 12 months		Cognitive (composite score) at 12 months		Receptive language (scaled score) at 12 months		Expressive language (scaled score) at 12 months		Cognitive (composite score) at 12 months	
		<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>
Contingent relevant responses	With no label	.012	.959	-.430	.058	-.054	.822	-.365	.113	-.413	.070	.041	.863
	With one label	.369	.109	.507	.023	.265	.258	.240	.308	.424	.063	.041	.862
	With more labels	.602	.005	.160	.500	.083	.727	.346	.135	.032	.894	.331	.154

Significant results are in bold.

Table 13

Pearson's correlation coefficients between the types of salient maternal response and BDIS-III receptive and expressive language scaled scores and cognitive composite score in ELGA and FT dyads, at 24 months (corrected age for ELGA infants).

		ELGA (<i>n</i> = 20)						FT (<i>n</i> = 18)					
		Receptive language (scaled score) at 24 months		Expressive language (scaled score) at 24 months		Cognitive (composite score) at 24 months		Receptive language (scaled score) at 24 months		Expressive language (scaled score) at 24 months		Cognitive (composite score) at 24 months	
		<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>
Contingent relevant responses	With no label	-.002	.995	-.273	.244	-.239	.310	-.118	.641	.112	.659	-.074	.771
	With one label	.219	.354	.262	.264	.189	.424	-.074	.770	-.134	.597	-.298	.230
	With more labels	.188	.426	.542	.014	-.222	.346	.007	.978	.126	.618	-.031	.903

Significant results are in bold.

Table 14

Linear regression of maternal contingent relevant responses with more labels at 12 months on BSID-III expressive language scaled score at 24 months.

	B	β	CI (B)	<i>p</i>
Maternal contingent relevant responses with more labels at 12 months	0.18	0.38	0.10-0.25	.019
Intercept	7.35		6.68-8.03	<.001

Significant results are in bold.

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