



Review Article

Sensorimotor contingencies in congenital hearing loss: The critical first nine months

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SUMMARY

In the recent two decades it became possible to compensate severe-to-profound hearing loss using cochlear implants (CIs). The data from implanted children demonstrate that hearing and language acquisition is well-possible within an early critical period of 3 years, however, the earlier the access to sound is provided, the better outcomes can be expected. While the clinical priority is providing deaf and hard of hearing children with access to spoken language through hearing aids and CIs as early as possible, for most deaf children this access is currently in the second or third year of life. We review the findings on neural development during the first year of life, including language development, multimodal interactions between sensory and motor systems, as well as brain connectivity. Some irreversible consequences of early auditory deprivation within the first year are exacerbated when the auditory system is not developed in synchrony with the other sensory and motor systems, incorporating hearing into other sensory and motor representation. The key role of the motor system (sensorimotor contingencies) in development of sensory representations is discussed. We propose that the first year includes critical developmental steps that should be exploited to provide the framework for optimal functional connectivity of language and cognitive networks.

1. Introduction

Thirty-two million children world-wide are deaf or hard of hearing (DHOH). Suggested best practice today is to provide hearing aids and cochlear implants within the critical period for therapy, ending near 3 years (Manrique et al., 1999; Niparko et al., 2010; Kral et al., 2019). Many professionals consider critical periods as a deadline to which we need to comply to, rather than a period of decreasing therapeutic potential with a final endpoint. Consequently, even in high-income countries where newborn hearing screening allows for early diagnosis of hearing loss, cochlear implant (CI) fitting typically does not occur until the second or third year of life, i.e. close to the end of the critical period (Kleijbergen et al., 2022). Current FDA criteria may contribute to it by defining the *minimum age* for cochlear implantation of 9-12 months (Lieu et al., 2020). Thus, cochlear implantation for those with severe to

profound hearing loss is uncommonly performed before 12 months of age (Mathew et al., 2021; Bruijnzeel et al., 2017).

Sales data from the three major CI companies (Fig. 1A,B), thus including the majority of all cochlear implantations globally, indeed demonstrate that less than 20% of profoundly deaf infants implanted within the first 3 years receive the CI in the first year. Even the countries with the highest proportion of implanted children in the first year, Australia and Germany, do not exceed 40% implanted in the first year. The proportion of the children implanted in the first year globally did not change from 2017 to 2024, nor did the proportion of children implanted in the second year change (one-way ANOVA, $F(6,68)=0.39$, $p=0.88$). Around 30% of these infants do not meet the expectations of the clinicians, and 50% of the variance remains unexplained (Boons et al., 2012). However, many studies demonstrated that effective hearing restoration before 9-12 months of age is beneficial for language

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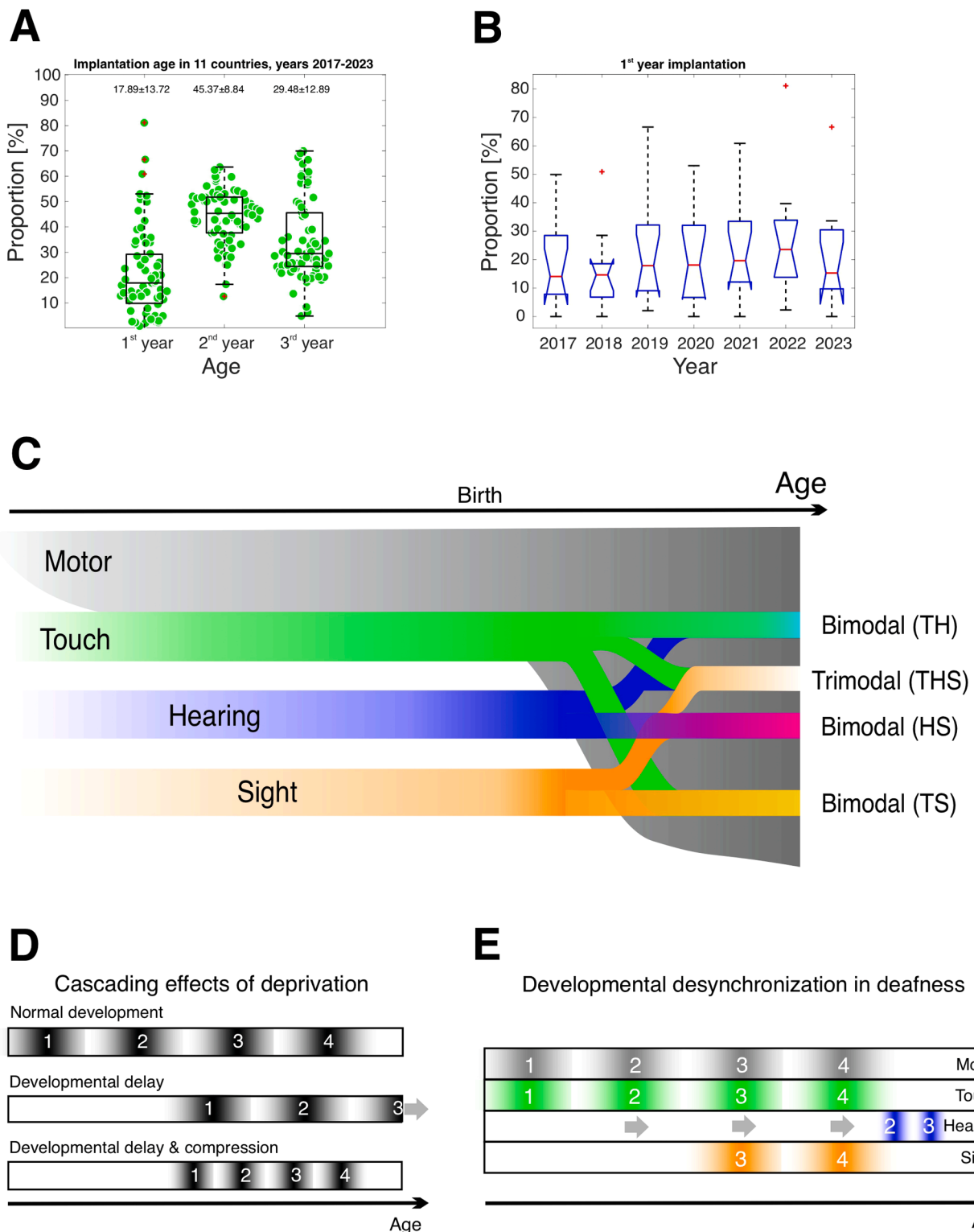


Fig. 1. A: Distribution of cochlear implants within children that obtained the first implant in the first 3 years of life (assumed perilingual deafness). Included are 11 countries: USA, Australia, Germany, UK, France, Spain, Brazil, Argentina, China, India, Sweden and years 2017-2023, representing the vast majority of globally implanted children. B: Change in the proportion of first year implantation over the years is not significant (one-way ANOVA, $F(6,68)=0.39$, $p=0.88$). C: Normal development includes an ordered sequence of developing motor and sensory organs (based on Stein et al., 2014). We suggest that this process happens against a background of motor development (grey color) that shapes sensory stimulation by motor action. T=touch; H=hearing; S=sight. D: Sensory deprivation with later sensory restoration cause a delay or a delay and a compression of the developmental sequence. Numbers correspond to different stages in the development of the given system, grey arrow indicates a shift of the developmental sequence. E: Under the assumption of a delay & compression, hearing loss during development desynchronizes a significant part of auditory development from sensorimotor development, and e.g. stage 3 coincides with stage 4 in the other systems. Grey arrows indicate the delay and compression of the auditory sequence.

acquisition (Ching et al., 2017; Hoff et al., 2019; Karltorp et al., 2020; Chweya et al., 2021; Ilg et al., 2024). Recently, it has been further shown that early communication, quantified by verbal turn-taking, promotes different aspects of brain development (Romeo et al., 2021; Romeo et al., 2018a; Romeo et al., 2018b). Hearing and communication, including prelinguistic communication, involves all sensory-motor modalities and executive function already in the first months of life. Thus, although sound can stimulate the brain as early as during intrauterine life, and may contribute to its maturation (Kral et al., 2016), it is active hearing and communicative interaction that provide critical input for brain development. As we argue here, the key developmental period of the first 9-12 months should not be passed without sufficient access to sound.

2. Multisensory and sensorimotor contingencies during development

A plethora of developmental processes take place simultaneously within the first months of life in a hearing child (Table 1). These include communication involving *multimodal inputs* (understood here as including motor and all sensory systems). The “cascading” processes during this period are dependent on each other (review in Lickliter, 2011). The pace of development and the co-dependence of the processes are influenced by the infants’ innate abilities, including near-sighted vision, limited ability to walk or crawl, grasp only what is at arm-length, absence of information on the meaning of sound and consequently limited representation of the environment. The gradual development of these abilities is essential for developing the representation of the outside world, much of which is driven by sensory inputs and their behavioral consequences. Gradual attainment of motor functions allows increasing the action radius incrementally and is itself

related to cognitive progress (Oudgenoeg-Paz et al., 2017) including executive functioning. Hearing is a part of this process, where all components need to act together in a developmental sequence so that the infant can build appropriate sensory representations and cognitive constructs of the environment (see below). These in turn serve as building blocks for higher-level cognitive skills such as language and communication.

The notion of a strong relationship between the developing body, multimodal (multisensory and motor) experiences and the development of language and cognition has been also addressed by the *embodied cognition theory* (Varela et al., 2016; Craighero, 2024). Language as a higher-order cognitive process depends on several sub-processes such as sensory perception, memory, attention, inhibition, and problem-solving skills. Therefore, spoken language development also depends on timely multimodal and cognitive mechanisms in a cascading developmental process (Fig. 1C-E).

The brain constructs a virtual internal model of the current environment (Johnson-Laird, 1983; Johnson-Laird & Byrne, 2002; Johnson-Laird, 2005) which combines information from many separate brain networks and is neurally defined by the actual brain state (Duncan, 2025). The metaphorical concept of such a “mental model” or a “cognitive map” refers the representations of the external environment (or imagination) at the given moment that humans use when they interact with the environment and systems within it (Furlough & Gillan, 2018; Duncan, 2025). An internal model includes all subjectively-relevant aspects of the environment that one is currently embedded in and the knowledge we have about the objects in it (Duncan, 2025). Such an internal model can be used to predict future events (Furlough & Gillan, 2018). Predictive processing (Friston, 2010) postulates that the active representations of the scene (context or mental model) provide predictions of sensory input that is compared to actual

Table 1

Development of the infant within the first 24 months. Shown are individual behavioral stages related to expected perceptual development and to communication changing from dyadic to triadic at 9 months. The transition at 9 months is considered critical by many studies (for review, see e.g. Tomasello et al., 2005). Sound discrimination ability is inborn (Cheour-Luhtanen et al., 1995; Kujala et al., 2004), but the ability for discrimination improves further with experience (Bishop et al., 2011). While data from the visual system suggest that categorial perception is achieved in the 4th month (Baillargeon & DeVos, 1991; Soska & Johnson, 2008; Soska et al., 2010), auditory studies suggest that these can be traced down to the first months of life already (Eimas et al., 1971; Jusczyk et al., 1983).

Age [mo.]	Behavior	Reference	Communication	Reference
0	Preference for mother language and voice	DeCasper & Fifer, 1980; Gervain et al., 2008; Mehler et al., 1988; Moon et al., 1993; Locke 1997	Dyadic	Egeren et al., 2001; Striano & Berlin, 2004; Bornstein et al., 2008; Striano & Rochat, 1999; Waxman et al., 1996
3	Cooing	Coplan & Gleason, 1990		
	Imitation of the caregiver - motherese	Snow and Hoefnagel-Höhle, 1977; Snow 1981; Kuhl et al. 1997; Kuhl 2000		
4	Categorization, sensory objects	Eimas et al., 1971; Jusczyk et al., 1983; Specke & Kenstenbaum, 1986; Baillargeon & DeVos 1991; Soska & Johnson, 2008; Soska et al., 2010		
6	Top-down interaction (visual)	Kouider et al., 2013; Emberson et al., 2015; Zhang et al., 2018		
	Canonical babbling (auditory)	Oller & Eilers, 1988; Oller et al., 1999; Juszyk et al., 2002		
7	Loss of discrimination of foreign phonemes	Werker et al., 1981; Werker & Tees, 1984; Werker & Tees, 1992		
9	Segmentation of speech stream into words	Ferry & Guellai, 2021	Triadic and beyond	Bruner, 1975; Carpenter et al., 1998; Striano & Rochat, 1999; Tomasello et al., 2005; Blythe et al., 2010
12	Object constancy (visual)	Xu & Carey, 1996; Xu et al., 1999		
	Word-level comprehension, semantics (auditory)	Golinkoff et al., 1987; Skeide & Friederici, 2016		
13	Lexicon built-up; Vocabulary spurt 13 - 24 months	Goldfield & Reznik, 1990; Segbers & Schroeder 2017		
15	50 words reached	Benedict 1979		

input. The resulting prediction error drives brain plasticity and provides information for an update of the inner model to re-match external world (Kral et al., 2017).

Newborns, however, have yet to develop the internal representations that serve as the foundation for constructing models of the external world. The formation of such representations solely from sensory input is a complex process influenced by numerous factors. For example, interpreting a visual scene from a specific viewpoint requires implicit knowledge of spatial organization, principles of perspective (variance of size depending on distance), object occlusion and reappearance with changes in viewing angle, and lighting effects like shadow formation. Because sensory input is inherently variable, and newborns lack prior knowledge about the structure of the world, the task of constructing an accurate internal model is *underdetermined*. In other words sensory input alone is too ambiguous to yield a stable and coherent representation of reality.

The elements that may instruct the development of an internal model of the environment and provide key a priori information are **sensorimotor contingencies** (O'Regan & Noë, 2001), defined as structured, lawful relationships (consistencies) between the organism's motor actions and the resulting sensory changes that occur due to these actions. In the above example, we might see our hands moving within the visual images and at the same time have neural information about the motor commands that moved the hands. Having information of the motor commands with proprioceptive feedback, together with the sensory impression of the movements we make, together with the soundscape caused by our motor act, allow to identify our action on the visual and auditory scene and thus deconstruct the scene. By that we can additionally fuse the individual sensory representations into one, higher-order internal model of the environment.

Sensorimotor contingencies thus allow to transform development of representations into a *determined process*: the brain not only possesses the information on how the infant moves its body (through proprioception, vestibular inputs, and efference copy of the motor program), but it also feels the contact with objects through touch (particular touching its own body) and seeing the moving body parts with the eyes. In addition, the child hears the sounds of objects manipulated with their hands, associating the caregivers voice with movement of the mouth with facial expressions in temporal synchrony. Sensorimotor contingencies, therefore, provide powerful cues for the developing brain to anchor and implement the rules that allow faithful sensory and subsequently high-level representations (Varela et al., 2016). Already in their first weeks of life infants follow active exploratory efforts rather than passive reflexive behavior (Gibson, 1988). Infants thus actively explore the environment. To support this, it is recommended to give them toys within reach, visual displays within eyesight, and correspondingly near-distance human interactions as well as salient auditory communication.

Information on sensorimotor contingencies indeed reaches the sensory cortex: Neural correlates of sensorimotor interactions have been described within the auditory cortex (Schneider & Mooney, 2018; Kral & Sharma, 2023). Consistent with the suggested role, secondary auditory areas have been implicated in action timing, which is disrupted by auditory deprivation (Cook et al., 2022; Hidalgo et al., 2020). Many developmental processes in the auditory cortex are considered "experience-expectant", shaped by genetic programming and experience (Kral et al., 2017). Even gene expression itself is experience-dependent through (epigenetic) modifications (Traniello & Robinson, 2021).

In the developmental sequence of the cerebral cortex, sensory areas are bottlenecks: they develop earliest in a defined developmental sequence (Fig. 1C) starting with touch, followed by hearing and then vision (cats: Stein et al., 2014; humans: Lickliter, 2011; Craighero, 2024). This sequence, which in humans begins in utero, ensures that the earlier developing systems can establish foundational processing without initial interference of the later developing systems. Only following this developmental step they can be mutually integrated together into multimodal representations. Touch, the

earliest-developing system, is uniquely interwoven with the motor system through direct connections between the somatosensory and primary motor cortices, along with parallel proprioceptive input to both regions (Gómez et al., 2021). At birth, all human sensory systems are rudimentarily functional and capable contributing to the formation of early multimodal representations (Craighero, 2024).

Multisensory interactions appear after the initial developmental steps in the sensory systems have been taken, and are critically shaped by sensory experience (Stein et al., 2014; Bean et al., 2022). Newborns can effectively learn to integrate different sensory inputs in the sensory cortex (Dall'Orso et al., 2020). Learning of multisensory interactions takes place in the first 3 months of life (Baillargeon & DeVos, 1991). Slow synaptic transmission (with long-duration postsynaptic potentials) in the juvenile brain (Aramakis et al., 2000) facilitates detection of roughly coincident inputs. The resulting strong excitation facilitates the "penetration" of sensory input deeply through the brain network of different cortical areas, up to the multisensory association areas. Active interaction assures the engagement of attention in learning (Seitz & Dinse, 2007). In early infancy, attribution of attention has been related to multisensory stimulation (Lickliter, 2011; Craighero, 2024). Thus, learning, attention, and multimodal integration are tightly linked.

3. Sensory features and sensory objects

Perception involves both continuous dimensions (such as pitch, loudness, and brightness which are perceptual correlates of sensory input) and categorical dimensions, as exemplified by speech perception, where small changes in specific acoustic cues can shift perception from one phoneme to another (McMurray, 2022). A well-studied example is the perception of voicing, a *distinctive feature* in phonology that distinguishes phonemes like /b/ and /p/. Although voicing is a binary phonological category (voiced vs. voiceless), it is signaled by the continuous acoustic cue of voice onset time (VOT). Neural processing must learn to map this continuous phonetic dimension onto categorical phonological representations, a task that is language-specific and develops over time. For instance, English and Spanish both use the voicing contrast, but they differ in where the categorical boundary for VOT is placed. Categorical perception is marked by discontinuities in the perceptual response to acoustic continua: listeners show heightened sensitivity across category boundaries and reduced sensitivity within them (Eimas et al., 1971). This phenomenon can be modeled within a multidimensional feature space as defining a boundary, where changes within the boundary (a category) produce the same percept, while crossing a boundary yields a new one. However, not all aspects of speech are perceived categorically; non-categorical perception also plays a role, even in spoken language (Apfelbaum et al., 2022; Rizzi & Bidelman, 2024). Thus, categorical and non-categorical perception together support the mapping from continuous sensory input to discrete linguistic representations (for review, see Chomsky and Halle, 1968; Stevens, 1998).

However, from a neuroscience perspective, "sensory" or "acoustic" features refer to the neurophysiological aspects of sound processing. Different acoustic features (like frequency, intensity, temporal modulation, frequency modulation, binaural relations and similar) are represented in the auditory system, often topologically (Schreiner & Winer, 2007). Such topological maps in the auditory system often represent continuous gradients and depend on experience with the given acoustic features (de Villers-Sidani & Merzenich, 2011; Weinberger, 2015). Sensory features can be grouped into a sensory object (category), defined as a neuronal representation of a delimited pattern of features that is subjects to figure-background distinction (Kral et al., 2017). Sensory objects are thus the result of grouping (categorization) of features into stable perceptual units (Bizley & Cohen, 2013). The grouping of speech sounds can be at different levels (in language e.g. phoneme, syllable, morpheme) and the critical level depends on behavioral context (Goldinger & Azuma, 2003). Categorization ability is linked to

additional behavioral consequences, like increased sensitivity to novel stimuli in a stimulus stream or similar responsiveness to clear and degraded features of the same stimulus.

To the best of our current knowledge, there is no empirical evidence supporting the existence of innate sensory categories in humans. Newborns are believed to acquire the capacity to categorize stimuli through experience and the active exploration of their surroundings. Consequently, sensory objects (categories) emerge after birth and are refined by sensory experience and active exploration of the environment. Categorization is closely linked to the behavioral significance of stimuli for an individual.

The newborn auditory brain has an innate capability to differentiate between sensory stimuli (Aslin & Pisoni, 1980; Cheour-Luhtanen et al., 1995; Kujala et al., 2004). This is an important precondition for subsequent grouping of stimuli into *different* meaningful sensory categories (or objects). Along with further improvement of refinement of feature sensitivity (Sanes & Woolley, 2011), the brain learns to determine features that are relevant and to ignore the nonrelevant ones, focusing on discriminating sounds of behavioral relevance. Thereby, sensory objects are established as representations extracted from multiple instances of a similar stimulus. Signs of categorical perception, reflecting the organization of sensory features into discrete perceptual sensory objects, and thus categorical perception, typically emerges within 1-6 months after birth, depending on task and sensory system (Eimas et al., 1971; Bailargeon & DeVos, 1991; Segal et al., 2016, see Table 1). By 4 and 6 months, infants also begin to construct three-dimensional representations of objects in their environment (Soska & Johnson, 2008). Consistent with the proposed role of sensorimotor contingencies in perception, motor development plays a cardinal role for the emergence of object-level representations (Soska et al., 2010).

Such object-level representation is also connected with appearance of Bayesian-type inference and top-down interactions. Top-down modulations (e.g. from object-level to feature-level) start shaping neuronal responses at the age of 6 months (Emberson et al., 2015). As a side-effect of developing categorial perception in spoken language, hearing infants stop differentiating phonemes by acoustic features that fall within one native phonetic category. It supports perceiving different realizations of the same native phonemes as the same sounds. This phenomenon appears at around 7-8 months after birth (Werker & Tees, 1984; Pons et al., 2009). A hearing infant can identify the same acoustic "label" (word) in speech signal in different contexts around 9 months of age (Ferry & Guellai, 2021; Bergelson & Swingley, 2015). Discrimination of speech sounds is likely further refined throughout the development in the sense of perceptual narrowing (Pons et al., 2009; Lewkowicz & Ghazanfar, 2009), crystallizing the phonological system as the vocabulary is increasing (during the second year of life). The first discriminations likely relate to linguistic categories larger than a phoneme (Nittrouer, 2006; McMurray et al., 2022), but articulation of the infant, and thus motor program, requires auditory feedback and a corresponding phonetic-level of representations.

Taken together, the development of object-level representation takes place within the first months of life, alongside with the emergence of the ability to categorize speech cues already at the age of 7-9 months of age.

4. Role of top-down control and the motor system

Categorization involves the individual calibration of representations by their subjective importance. Historic evidence shows that the experience shaping this process must be active to assure that sensorimotor contingencies can instruct the emerging sensory representations (Held & Hein, 1963; Levi & Li, 2009). Sensorimotor abilities of infants open and close environments for learning (Smith et al., 2018), consistent with brain development: already in the first year of life, active auditory experience is more effective in shaping sensory responses compared to passive exposure (Benasich et al., 2014). For hearing infants, active communication with their caregivers is influencing later development

(Goldstein et al., 2009; Feldman, 2007). Even in infants with intact sensory organs, the quantity and quality of adult-infant interactions extensively affect language development and language-related cortical development (Romeo et al., 2018a; Levin-Asher et al., 2022; Kuhl, 2004). It is, therefore, the combined effect of early access to sound and active listening within a communication context (involving sensorimotor integration) that determines the development of age-appropriate cognitive abilities and language (Kuhl, 2011; Kuhl, 2004). It has been argued that the active communication with a partner (social interaction) is a key factor that promotes plasticity and thus permits language learning (Kuhl, 2011).

Correspondingly to the extensive behavioral development in the first 9 months of life, the development of long-range functional connectivity in the brain accelerates within the first year (Damaraju et al., 2014), integrating the sensory systems into the large-scale networks (e.g. default mode network) from 6 months on (Gao et al., 2017). Hemispheric asymmetry of language representation appears already at 11.5 months (Emerson et al., 2016). Functional connectivity analyses suggest that the newborn brain follows the architecture of a small-world-network (with predominantly local connectivity and less abundant long-distance connectivity), with further strengthening of long-range connections from ~1 year on (Gao et al., 2017). In the auditory cortex, the basic building block of the neocortex, the cortical column, starts gradually functioning after birth (Kral et al., 2005), with the increasing ability to integrate top-down influences into afferent processing (Kral et al., 2006; Yusuf et al., 2022). Hearing experience is fundamental for development of such functional interareal connectivity in the auditory cortex (Yusuf et al., 2021) but also for local (intrinsic, within-area) connections between supragranular and infragranular layers (Yusuf et al., 2022). Hearing experience is fundamental for the high-to-low frequency coupling of oscillatory activity (Yusuf et al., 2017), a precondition for integration of top-down inputs into sensory processing (Yusuf et al., 2024).

Top-down modulation is also the route by which the motor system can affect auditory processing. We argue that the motor system has a key role in the development and provides a priori information for the sensory systems that can be used in processing sensory inputs during active exploratory behavior: the motor command that is causing a movement predicts the proprioceptive feedback, but through the internal model of the environment may predict also the somatosensory, visual and auditory input. As mentioned above, this is reflected in predictive processing, an influential theory of learning and perception (Rescorla & Wagner, 1972; Rescorla & Solomon, 1967; Friston, 2010). This theory has been extensively verified by many learning paradigms.

The auditory system has a tight relation to the motor system, given that motor actions, even independent of vocalizations, frequently generate sounds that can be related to the motor act (Lemaitre et al., 2018). Motor feedback does indeed influence neuronal activity in the auditory cortex: motor activity modulates responses in the auditory cortex (Gale et al., 2021; Bagur et al., 2018; Schneider & Mooney, 2018). With respect to spoken language, sensory representation may be complemented by information from proprioceptive feedback from articulatory apparatus and the motor representation of vocal tract movements during vocal production (Liberman & Mattingly, 1985). Motor activity and the proprioceptive feedback can be compared to the sensory input resulting from it, and may serve as information for decoding sensory input. Consistent with these concepts, particularly predictive processing (where the brain also predicts what the vocal movement should sound like), the responses to the own voice are suppressed in the auditory cortex (Eliades & Wang, 2017; Numminen et al., 1999). A reciprocal link between motor and sensory representation explains the activation of the motor cortex representing vocal tract during passive listening to speech (Murakami et al., 2012; Cheung et al., 2016; Nourski et al., 2023) and activity in voice areas during silent reading (Perrone-Bertolotti et al., 2012). Infants at 4.5 months demonstrate the association between movements of the mouth, visual input and the speech heard (Yeung &

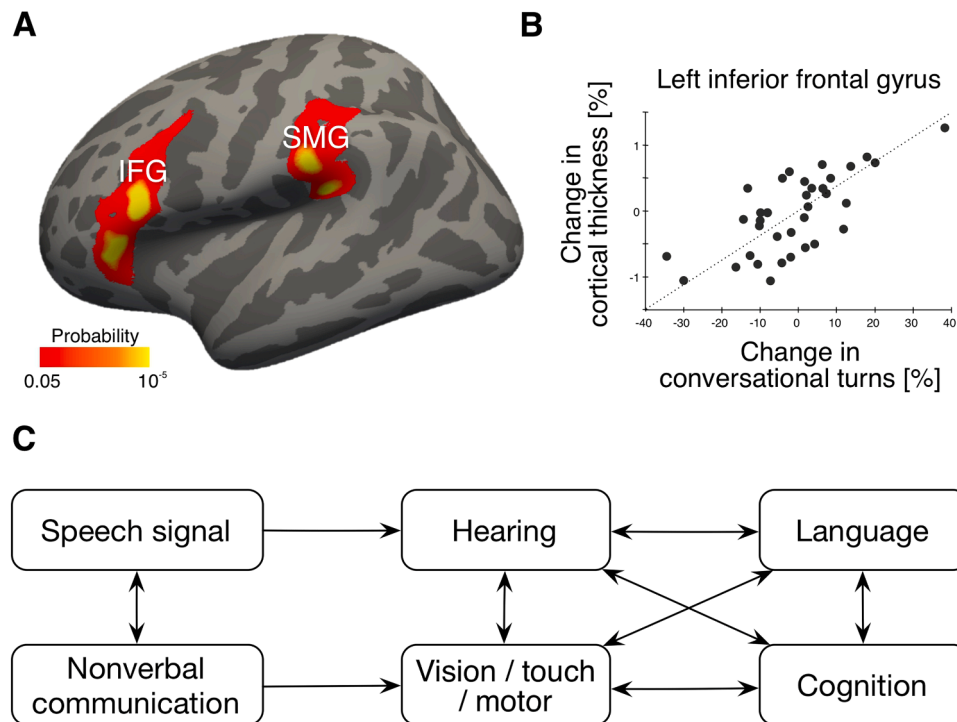


Fig. 3. A: Intervention to increase adult-child conversational turns slightly but significantly increases cortical thickness in language-relevant regions of left supramarginal gyrus (SMG) and left inferior frontal gyrus (IFG), and causes stronger connectivity between these areas. Reprinted with permission from (Romeo et al., 2021). B: Effect of increased conversational turns on cortical thickness in inferior frontal gyrus, data pooled over all studied groups. Dashed line shows the diagonal. C: Schematic illustration of the effect of total communication on development of language and cognition. While in the absence of hearing, the multimodal influence between sensory systems cannot take place, there is a direct effect of non-verbal communication on language and cognition.

motor systems provide a window of opportunity – or vulnerability - for these processes.

Evidence suggests that language acquisition requires both early exposure and active interaction; for example, passive video watching is not sufficient for language acquisition (Kuhl et al., 2003; DeLoache et al., 2010). At least ~10 hours of weekly active spoken language communication are required for hearing children of deaf parents to acquire spoken language (Schiff-Myers, 1993). Thus, language learning appears to be “gated” by socially-contingent interactive contexts (Lytle et al., 2018; Kuhl et al., 2003; Bosseler et al., 2024). Additionally, infants’ active manipulation of objects has been found critical for the development of vocabulary (Slone et al., 2019). Developmental absence of active social interaction leads to severe linguistic and cognitive deficits in adulthood (Nelson et al., 2013; Nelson & Gabard-Durnam, 2020). These deficits are not fully reversible if the social deprivation lasts longer than 6 months after birth (Sonuga-Barke et al., 2017; Thompson & Steinbeis, 2020). While it is often implicitly assumed that prefrontal cortex and executive functions develop late, cardinal steps in cognitive development are already outlined within the first months of infant life (Rose et al., 2005; Fiske & Holmboe, 2019). Development of some foundational executive functions is subject to an early sensitive period of the first 6-12 months (Thompson & Steinbeis, 2020), and this partially explains why restoration of auditory input beyond this period does not guarantee restoration of normal cognitive function (Kral et al., 2016), including relative risk for underperformance in conceptual learning, attention, sequential processing, and factual and working memory (Kronenberger et al., 2014). Many cognitive and executive functions develop later, but the interaction between cognition and sensory representation is established early and active communication with a social partner is key for this process (Bosseler et al., 2024).

6. Deaf and hard of hearing infants

So far, we have elaborated the importance of multimodal input for the development of hearing infants. Typical multimodal and sensorimotor synchrony offers a theoretical framework for understanding the large unexplained variance in speech comprehension if access to sound is not initiated in the first year of life (Duchesne et al., 2009; Niparko et al., 2010). Specifically, the unaccounted variance in the spoken language delay of DHOH infants may be related to disrupted multimodal auditory because of the hearing loss. In the absence or reduction of auditory input, the infant relies mostly on visual information and touch for synchronization of behaviors. However, vision cannot replace hearing: vision and hearing are fundamentally different and inform the brain on the environment in differing but complementary ways. For instance, while vision is organized around spatial aspects, hearing is organized around timing, with corresponding auditory dominance in perception and in multimodal training when discrepancies of the sensory input between the two modalities are present (Barakat et al., 2015; McGovern et al., 2016; Amadeo et al., 2019). Thus, early deafness is related to impaired performance in the time domain in other modalities, including fine motor coordination (Horn et al., 2006), sensorimotor transformations (Li et al., 2022) and serial sensory processing (Rinaldi et al., 2018). Spoken language fundamentally depends on precisely timed features that unveil across varying timescales, which are minimally provided by the spared sensory systems cannot provide. In addition, early auditory deprivation affects brain organization, such as, partial recruitment of the auditory areas by the visual processing (Nishimura et al., 1999; Petitto et al., 2000; Lomber et al., 2010; Hauthal et al., 2014). Such crossmodal reorganization precludes audiovisual interactions in the secondary auditory cortex and visual cortex (Land et al., 2016). Indeed, deaf children exhibit impairments in bimodal interactions, leading to a greater reliance on other sensory systems (Schorr et al., 2005; Corina et al., 2022; Kral & Sharma, 2023). Thus, neuronal

interactions between sensory systems are affected by congenital deafness.

Hearing loss influences the behavioral patterns of both the caregiver and the infant in dyadic and triadic interactions (Cejas et al., 2014; Fagan et al., 2014; Quittner et al., 2013; Cruz et al., 2013; DesJardin et al., 2009). On average, hearing mothers of severe-to-profound DHOH children tend to be more controlling in their verbal and nonverbal interactions (Fagan et al., 2014), spend less time in coordinated joint attention with the child (Cejas et al., 2014), use less complex spoken language in terms of mean length of utterance (Goldin-Meadow & Saltzman, 2000; Fagan et al., 2014), and use more directive spoken language compared to spoken language directed at hearing infants (Fagan et al., 2014; Ambrose et al., 2015). By nine-months of age, infants with hearing loss less reliably and actively elicit their mothers' attention by means of smiling, greeting, or reaching and demonstrate more self-comforting and repetitious motor behaviors (Kuhl, 2007) compared to typically-developing infants (Koester, 1995). At 12-13 months of age, they produce fewer topic-initiating behaviors than typically developing infants at the same age (Cejas et al., 2014). At the age of 10 - 20 months they show reduced involvement in book-reading interactions (Zaidman-Zait & Dromi, 2007). If hearing restoration happens after these periods, infants' interaction with caregiver has already been significantly affected, and can impact how hearing becomes embedded into neural systems.

From a neuroscience perspective, brain development exhibits sensitive periods during which the development of the sensory system is more susceptible to experience (Nelson & Gabard-Durnam, 2020; Kral et al., 2019). Auditory behavior will be permanently impaired if experience fails to occur during a critical period, with only limited recovery possible (review in Kral, 2013). In deaf animals who received chronic cochlear implant stimulation, developmental maturation of the auditory system involved active paradigms and approximated a natural environment

(Klinke et al., 1999; Kral et al., 2002; Fallon et al., 2009; Kral et al., 2013a), making it possible to exploit sensorimotor contingencies discussed above. Auditory critical periods were similarly observed in animal models and DHOH children, where absence of hearing could not be sufficiently compensated with CIs at later age (children: Manrique et al., 1999; Gordon et al., 2015, animal models: Kral, 2013). This is because early auditory experience extensively shapes the circuits in the cerebral cortex (Kral & Sharma, 2012) (Fig. 4) and thus all plasticity in later life is dependent on the circuit adaptation induced during the sensitive periods. It is interesting that development of cortical synapses happens in hearing children within the first 12 months of age, and if development in congenital deafness is extrapolated from the cat model to human, while it is delayed, synaptogenesis is still taking place within the first 12 months (Fig. 4). This suggests that the initial cortical network structure and function is laid down within the period that currently fails to be exploited clinically in deaf children.

While the initial developmental learning process is likely related to the ability of neuronal networks to sculpt features in a process similar to connectionists' deep learning neuronal networks, the subsequent steps are multilevel with a hierarchy of object processing – from phonological objects to morphemes and grammar that develop in a cascading process (review in Skeide & Friederici, 2016). The subsequent steps of sensory and cognitive development are tightly related. Intervention that is delayed (i.e., after this period has been missed), desynchronizes the developmental cascade (Fig. 1E) with an impact on the maturation of sensorimotor functions.

Interestingly, primary sensory cortical areas follow a faster developmental sequence than higher-order areas (Elston et al., 2009). Indeed, the critical period for the auditory development under cochlear implant stimulation has been observed in the primary auditory cortex (Kral et al., 2002; Kral et al., 2013b). Missing the critical period affects the corticocortical circuitry (Yusuf et al., 2017) and top-down interactions (Yusuf et al., 2021). The timing of the critical period for cochlear implantation in prelingually deaf children is strongly related to early auditory areas, as shown by P1-component of the evoked potential (Sharma et al., 2002; Sharma et al., 2005) generated in the auditory cortex rather than language and associative areas. Thus, the initial bottlenecks in development are the sensory areas, pre-processing acoustic features and supporting the categorizing sounds into objects and feeding with object-level representations the higher-order areas and cognition. Key developmental steps in these areas are again taken within the first 12 months of life.

Untreated pediatric hearing loss puts cognitive functions at risk (Kronenberger et al., 2014; Marschark et al., 2017; Davidson et al., 2019; Kral et al., 2016). This does not mean that cognitive performance is affected in each DHOH child per se; some of DHOH children perform as well as the best hearing children. But individual cognitive sub-functions are at risk, which may result in larger intracognitive variance in some of DHOH children, and thus more borderline and problematic outcomes in individual subfunctions in some DHOH children. Untreated auditory deprivation influences the synchrony of caregiver-infant interactions, and can result in reduced and/or slower infant responses of the DHOH infant to the interlocutor, and eventually to less sequential relations and prolonged lag time of responses during interaction. This may result in less or qualitatively reduced input to the infant, less mutual attention supporting conceptual development and reduced ability of the DHOH infant to participate as both initiator and receiver of communicative interaction. Consequently, sensory dyssynchronization in the sensitive period for social and language acquisition will be associated with reduced receptive and expressive language acquisition and cognitive abilities (Bornstein, 1989).

7. Intervention in DHOH infants within first months of life: open questions

For DHOH infants, access to sound via hearing aids can be achieved

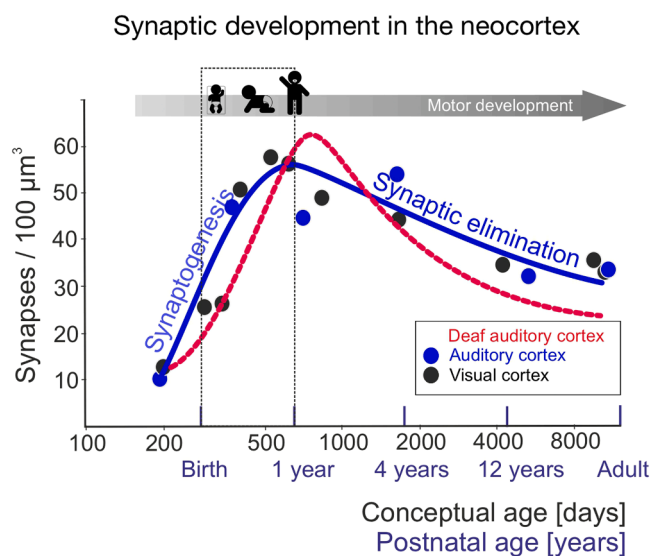


Fig. 4. Synaptic development in the human cortex, solid circles represent data from (Huttenlocher & Dabholkar, 1997). Trajectory of hearing children (blue line) approximated from visual and auditory development in children. Deaf development (red dashed line) is unknown in humans and has been extrapolated using (Workman et al., 2013) from congenitally deaf cats (Kral & Sharma, 2012) (conceptual age of humans: 280 days; cats: 64 days). Initial synaptic organization is established in both group of infants within the first 12 months. Pictograms approximate motor development, dashed rectangle the first 12 months of postnatal life. Synaptogenesis overlaps with the immature motor system, sensorimotor interactions are thus limited to near space. One consequence of the alteration of developmental sequence is a change in function of the cortical networks that are incapable of incorporating top-down auditory interactions (Berger et al., 2017; Yusuf et al., 2022).

within the first 3 months (Wood et al., 2015). For those who cannot benefit from hearing aids, cochlear implantation is a viable solution which is technically possible below 6 months of age, provided a child is medically fit and appropriate infrastructure is available (Lesinski-Schiedat et al., 2004; Waltzman & Roland, 2005; Tait et al., 2007). This timeline would substantially improve access to sound through HA and CIs before 9 months, with a known positive effect on language acquisition (Leigh et al., 2013; Ching et al., 2017; Hoff et al., 2019; Karltorp et al., 2020). The age limit for cochlear implantation is ultimately a surgical decision. Many clinicians rely on behavioral confirmation of objective measures of hearing. This is possible even before the age of 6 months, provided that the follow-up of the child is efficient, the team is experienced and there are no delays in care. Suggestions of 1-3-6 months pattern (identification-diagnosis-intervention) for DHOH children have been put forward by other authors as well (Findlen et al., 2023). We want to encourage the improvement of the efficacy of the follow-ups to allow such early interventions, also to allow further studies to strengthen the evidence-base for such early intervention.

One difficulty is the tracing of infants' developmental trajectory during sensory and cognitive development. Testing of the linguistic development of these children represents a challenge, as it is difficult to separate the interwoven sensory, linguistic, and other cognitive development. Checklists such as Auditory Skills Checklist (Meinzen-Derr et al., 2007) and parent-report measures such as Functional Listening Index (Coninx et al., 2009; Ching & Hill, 2007; Davis et al., 2022) may provide additional information of the earliest auditory, listening and vocal development of children before reliable cognitive and linguistic testing. Such measures have provided evidence of better outcomes with intervention below 9 months of age (Culbertson et al., 2022). However, the relation of these and standard linguistic measures require more attention in the future to allow individualized approach to early hearing loss.

From what has been reviewed here, development of multimodal representation is critical during the first months of life (Havy et al., 2017). Communication is more than passive sensory perception: it involves active exploratory efforts of the child. Infants are not passive recipients of sensory input, but rather actively search for interaction already within the first 5-12 months of life (Goldstein et al., 2009; Striano & Bertin, 2004; Gibson, 1988). Sensorimotor system plays a mediating role between communicative experiences and language development (Salo et al., 2022). However, more research is required to understand whether and how earlier access to sound may beneficially influence the downstream cognitive consequences of hearing loss.

In the light of these considerations, it is problematic that the **communication within the family** remains a **blind spot** in auditory intervention. In the clinical setting, it is unclear how much and what quality of communication is provided to the DHOH child. DHOH children require caregivers who provide them a rich language environment ("super communicators", Aragon & Yoshinaga-Itano, 2012). Caregiver language input is a critical feature during the early life particularly for DHOH children (Nittrouer et al., 2020; Holt et al., 2022). Caregiver's sensitivity and education are among the strongest factors affecting the outcome of intervention in DHOH children (Quittner et al., 2013; Cejas et al., 2018). The structure of linguistic input, particularly the active verbs, are another important factor for language acquisition (Warner-Czyz et al., 2024). Thus, there is a need for monitoring and guiding communication in the family. Technology allows quantifying the amount of interactions, e.g. using the Language ENvironment Analysis system (LENA) that provides data regarding the number of vocal interactions (turn-taking) between the infant and its caregivers, the number of words produced by an adult in the vicinity of the infant, and the count of the infant's vocalizations, extracted from up to 16 hours of recording in the infant's natural environment. LENA data help caregivers to reflect upon what they are doing in everyday settings and discover for themselves goals and strategies (Ganek et al., 2018;

Levin-Asher et al., 2022; Aragon & Yoshinaga-Itano, 2012; Kondaurova et al., 2022). While CIs and hearing aids are increasingly equipped with such technology, additional technologies are needed for other aspects of communication.

Ultimately, cochlear implantation within the critical period for hearing development shows that the brain is sufficiently flexible to allow a somewhat later initiation of hearing. Nonetheless, because plasticity shows a decreasing gradient within this period, earlier intervention is generally better. For further improving the outcomes of infants born with hearing loss we need to (i) raise awareness among otolaryngologists, neurologists, pediatricians, about consequences of hearing loss within the first months in life; (ii) initiate further studies on early interventions, comparing outcomes of children with improved access to sound within the first 9 months of life to those with later interventions, (iii) quantify listening skills also at stages when language scores are not available, (iv) quantify and analyze multiple domains of communication (including, but not limited to turn taking, vocalization and multimodal gestures in early infancy), (v) develop systems for automatic coding of communication including gestures and the input from the caregiver, (vi) combine these data with physiological brain imaging results and (vii) combine these data with quantified cognitive abilities of the infant.

8. Conclusion

The developing brain critically depends on sensorimotor contingencies through active exploration and communication starting at birth. Sensory deprivation in one modality can result in its developmental desynchronization from the other modalities involved in active communication. In the case of congenital hearing loss, we argue that the current habilitation practice is too late. The cumulative evidence supports the restoration of hearing at the earliest opportunity after birth in conjunction with a caregiver-led infant-directed communication, before the milestone of 9 months of age has been reached.

CRedit authorship contribution statement

Andrej Kral: Writing – review & editing, Writing – original draft, Visualization, Resources, Funding acquisition, Formal analysis, Data curation, Conceptualization. **Liat Kishon-Rabin:** Writing – review & editing, Writing – original draft, Funding acquisition, Conceptualization. **Gerard M. O'Donoghue:** Writing – review & editing, Writing – original draft, Resources, Conceptualization. **Rachel R. Romeo:** Writing – review & editing, Writing – original draft, Resources, Conceptualization.

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Data availability

Data will be made available on request.

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