

Implications of developmental plasticity for the language acquisition of deaf children with cochlear implants

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Abstract

The study of language acquisition in profoundly deaf children with cochlear implants informs us about the developmental plasticity of the auditory system. Sensory activity leads to neural development, and the sustained effects of sensory inactivity can lead to a loss of responsiveness. These effects may be reversed by the subsequent provision of sensory stimulation, such as that delivered by cochlear implants. Behavioral and electrophysiological research on the effects of speech deprivation on language acquisition shows that the age and modality of language acquisition is an important determinant of adult linguistic performance. Studies on profoundly deaf children deprived of speech stimulation, and then provided with a cochlear implant giving them access to the speech frequencies, shows that congenitally deaf children implanted under the age of around 5 years are likely to perform better on speech perception and speech production tasks than children implanted at an older age. Further investigation is required to understand why these large individual differences exist. In addition, other key issues for research are the effects of compensatory visual and somatosensory development prior to implantation, whether there is a maturational delay that approximates to the period of speech deprivation prior to implantation, and whether there are a number of sensitive periods that together describe the cascade of processes that underlies language acquisition. © 1998 Published by Elsevier Science Ireland Ltd. All rights reserved.

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1. Developmental plasticity and its physiological basis

1.1. Overview

This review argues that there are three major tenets of developmental plasticity. First, sensory

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activity leads to neural development, and this is often known as the 'glow and grow' theory. Second, the sustained effects of inactivity (sensory deprivation) can lead to a loss of responsiveness and selectivity in the auditory system. Third, the effects of inactivity may be reversed by the subsequent provision of sensory stimulation, and this is related to studies showing the protective effect of electrical stimulation by means of a cochlear implant.

1.2. Sensory activity leads to neural development

The constructive role of activity in brain growth has been succinctly and persuasively reviewed by Purves [1], where he has marshaled data principally within the somatosensory and visual domains to show that activity levels modulate differential growth patterns in sensory cortex. King and Moore [2] have recently reviewed the auditory data, and drawn a similar conclusion. They argue that sensory activity modulates the development of cochleotopically organized maps in the mid-brain and cortex. The combined weight of the data indicate that the capacity for change in the developing system is greater in the immature than in the mature system.

Purves [1] further suggests that the sensitive period observed in psychological studies of language acquisition may be explained by a progressive weakening of the link between activity and growth in maturing mammals. That link, however, does not disappear at adulthood; indeed, recent evidence shows that the learning process continues throughout the life cycle [2–4].

1.3. Sensory inactivity or sensory deprivation and compensation

The effects of sensory deprivation are well known and have been documented in the visual and somatosensory systems (e.g. [5,6]). Much of the work on the animal auditory system has been based on neonatal cochlear ablation which is known to lead to major anatomical reorganization (e.g. [7,8]). Another approach has been neonatal deafening at the high frequencies by ototoxic drugs (e.g. [9]), which is known to lead to

decreased cochleotopic representation of those high frequencies in the midbrain and cortical regions.

Investigations using tomography and electrophysiological measurement techniques (e.g. [10–12]) have shown that the longer the duration of profound to total deafness, the lower the level of auditory cortical activity. For example, Ito et al. [10] investigated the level of auditory cortical activity in nine totally deaf patients using positron emission tomography, and found that activity associated with acoustic stimulation decreased with increasing duration of deafness. Pelizzone et al. [11] bilaterally implanted a deaf patient, where one ear was congenitally deaf and the other had acquired profound deafness at 7 years of age. Implantation of the congenitally deaf ear at the age of 57 years resulted in clear sound perception, but no speech perception. Three years later the acquired deaf ear was implanted, enabling the patient to understand speech without lip-reading. Using long latency responses and magnetoencephalography, Pelizzone and his colleagues observed lesser activity in response to 1 kHz tone bursts in the congenitally deafened ear.

Jordan et al. [12] have recently measured the late auditory potentials in five cochlear-implant patients followed up over 6 months. Initially, the feature known as N1, normally found about 100 ms after stimulus offset, occurred later in implant patients, but with more experience with the implant, the latency approached normal values. In two postlingually deafened patients, this effect was marked (age at implantation 16 and 49 years, duration of deafness 13 and 0.3 years, respectively). The effect was weaker and more variable in the three prelingually deafened patients (age at implantation 16, 17 and 32 years, duration of deafness 16, 15.5 and 31 years, respectively). Hence, the data indicate the effects of age at onset of deafness (pre- versus postlingual), and of duration of deafness. The effect of age at onset of profound deafness is further considered in Section 2 of this review.

Another determinant of developmental plasticity is the mode of stimulation. Development occurs in a multi-sensory environment, and in the circumstances of auditory deprivation, compensa-

tory plasticity occurs where increased representation and activity may be found in visual and somatosensory cortex (see [13] for a recent review). An early example of this work may be found in Wolff and Thatcher [14], who performed electroencephalographic topographic mapping in 79 severe and profoundly deaf children and also in a matched group of hearing children. Electroencephalographic coherence and phase, which reflect patterns of cortical connectivity, was measured in the resting eyes-closed condition. Wolff and Thatcher found that deaf children showed less evidence of connectivity in the left auditory cortex than hearing children, and that deaf children showed greater evidence of connectivity in the right visual cortex which suggested compensatory plasticity.

1.4. Sensory deprivation may be reversed

Ponton et al. [15] have reported preliminary findings that show restoration of auditory function in deaf children as a result of cochlear implantation. In addition, they suggest that maturational delay in implanted children approximates the period of auditory deprivation prior to implantation. They measured late auditory potentials in response to biphasic electrical pulses in six implanted children, and late auditory potentials in response to acoustic click stimuli in eight normal-hearing children. They found that the latency changes for the P1 component had the same rate of maturation in both groups of children. This result was obtained in a cross-sectional analysis, and it is important to obtain longitudinal data. Nevertheless, it is clear that the auditory system does not mature without stimulation, and that it retains its plasticity during the period of profound deafness. Following cochlear implantation, the normal maturational sequence appears to occur, albeit delayed.

Few other investigators have examined a loss that is reversible or temporary in the developing auditory system. Early exceptions are the work of Stein and Schuckman [16] and Knudsen et al. [17]. Stein and Schuckman plugged the external meatus of neonatal rats for a minimum of 32 days, leading to a temporary conductive loss. They subsequently trained the rats to respond to direct electrical

stimulation of the primary auditory cortex, and found that experimental animals showed poorer auditory learning compared with control animals. Ear plugging of adult rats did not lead to similar effects. Knudsen et al. [17] studied the effect of an ear plug on the auditory localization of noise bursts in the barn owl. When the conductive loss occurred during development, owls were able to adjust to the effect of the ear plug, and localization performance was restored. On removal of the ear plug, owls were able to readjust, provided this occurred within 200 days following birth [18]. A similar experimental design using adult owls did not lead to similar effects. Hence, both the Stein and Schuckman [16] and Knudsen et al. [17] studies show a difference in degree of neural plasticity as a function of maturity.

The research on the protective role of chronic electrical stimulation in the absence of acoustic activity is related to the literature on reversing sensory deprivation. Chouard et al. [19] initially showed that chronic electrical stimulation partially prevented atrophy in the auditory brainstem which had been denied normal acoustic-afferent stimulation in the neonatally deafened guinea pig. Subsequently, Matsushima et al. [20] subjected the implanted cochleas in kitten to long-term electrical stimulation using biphasic pulses at 16 h/day over a period of 3–4 months. They found that cell somata areas in the kitten auditory brainstem were larger on the electrically stimulated side rather than on the non-stimulated control side denied acoustic stimulation from its afferents 4 months after deafening by ototoxic drugs. Lustig et al. [21] used kittens that had been neonatally deafened by ototoxic drugs, and implanted at 6–16 weeks of age. The animals were then electrically stimulated with biphasic pulses for 4 h/day, 5 days a week over a period of 3–6 months and sacrificed at around 8 months of age. They reported decreased shrinkage in the auditory brainstem which had been stimulated by its afferents following electrical stimulation of the spiral ganglion cells. Taken together, these and other studies indicate that chronic electrical stimulation at the low levels typically delivered by a cochlear implant has a protective effect on the developing auditory system [22].

2. Language acquisition and developmental plasticity

The recent electrophysiological literature briefly reviewed in Section 1 shows that the brain continues to adapt and reorganize in response to environmental change. The following section discusses the physiological evidence in the light of the literature on language acquisition and associated sensitive periods. The reader is also referred to two recent reviews which provide a broadly physiological [22] and a rehabilitative [23] perspective.

Lenneberg [24] made a major contribution to the literature by suggesting that biological considerations were necessary for the understanding of behavior, with particular reference to language acquisition. He proposed that there was a critical period for language acquisition that occurred between the age of 2 years and adolescence, and that its termination was related to loss of adaptability and inability for reorganization in the brain. Although Lenneberg was incorrect in assuming the critical nature of the sensitive period, and the inability for the adult brain to reorganize, his basic premise emphasized the importance of biological considerations for language acquisition and this remains unchallenged (in their reviews, Kyle [25], Ruben and Rapin [26] and Neville [13] draw a similar conclusion).

At the time, Lenneberg [24] had little direct evidence. He reviewed studies on the language acquisition of child and adult patients recovering from traumatic aphasia, which show that children are more likely to relearn language than adults. Lenneberg acknowledged that an overall evaluation of this literature was difficult because the research tended to be case studies or very small series of patients. He concluded that if aphasia strikes before puberty, the child will reacquire language, albeit with a delay of around 1 year. If, however, aphasia strikes afterward, he stated that children can regain language, but that there would be odd hesitations, pauses, searching for words and inappropriate sound or word sequences. However, in a recent review, van Hout [27] has suggested that recovery is not complete even in children who have been aphasic from an early age, and that the recent literature indicates greater

similarities between children and adults who are aphasic. For example, although aphasic children may be able to speak, tests of language by eye are affected.

2.1. Language deprivation

The results of the so-called isolation studies in language acquisition parallel those of the sensory deprivation studies, and provide qualified support for Lenneberg's [24] hypothesis of a sensitive period for language acquisition. The qualification is that although the isolation studies are direct and poignant, they remain case studies. The studies show that the acquisition of language is relatively robust, in that recovery occurs even after many years of isolation. However, should isolation persist until adolescence, then language acquisition becomes more difficult. Two examples of this literature are Mason [28], who reported on a child isolated in a dark room for 6 years with her deaf mother. Within 18 months of discovery, the child could read, write and compose stories, and had a vocabulary of over 2000 words. In contrast, we have the well-known example of Genie [29] who was discovered at 13 years of age. She had been isolated since the age of 20 months. Even after 7 years of rehabilitation, Genie never achieved good language acquisition. However, it is also possible that Genie had additional learning disability resulting from the unusual conditions in which she had been raised.

Newport and her colleagues have studied language acquisition shown by congenitally deafened adults [30]. One group of adults had learned to sign from birth, having had deaf signing parents; the second group learned sign language at school age entry (4–6 years of age); the third group were adolescent learners of sign language. All of the deaf adults had signing experience for at least 40 years, and they were tested on their production and comprehension of sign language. Newport and her colleagues found that those adults who had learned to sign from birth scored better than adults who learned to sign at school age entry, and those adults scored better than the adults who had learned sign language during adolescence. Johnson and Newport [31] extended the

work to hearing adults and investigated the age at second language acquisition. They studied 46 Chinese and Korean immigrants in the United States who had never spoken English previously, and who had arrived in the US at ages ranging from 3 to 39 years. The immigrants listened to spoken sentences, where half of the sentences were grammatically incorrect. Johnson and Newport found that the age of acquisition of the second language determined grammatical performance. The seven immigrants who had arrived at the ages 3–7 years performed as well as 23 native speakers of English on a test of English grammar, but those who arrived at later ages performed progressively worse with increasing age at arrival. The results of both Newport studies demonstrate that the age and modality of language acquisition is an important determinant of performance.

Investigators using electrophysiological techniques have come to similar conclusions (see, for example, the review by Neville [13]). One example of this work is Neville and Lawson [32], who compared event-related brain potentials to peripheral and central visual stimuli in three groups of subjects. The first group were congenitally deaf adults whose first language was sign, the second were normal hearing adults born to deaf parents, and whose first language was sign, and the third group were normal hearing adults. Evoked response potentials were measured in response to visual stimuli presented in either the left or right visual field. Increased activity in the left hemisphere was found in hearing adults compared with deaf and hearing signers. In contrast, deaf adults showed increased activity in visual cortex of both hemispheres which suggested neural compensation. Furthermore, Neville and Lawson found systematic asymmetries in the visual detection of motion. In deaf and hearing signers, they found a right visual field advantage, whereas in hearing adults they found a left visual field advantage. They concluded that this pattern of results indicated that the age and modality of early language experience has a marked effect on the pattern of hemispheric specialization.

2.2. Reversing auditory language deprivation through cochlear implants

Lenneberg's [24] hypothesis of language acquisition is directly tested by the investigation of deaf children with cochlear implants. Children who are provided with these devices are profoundly or totally deaf; they have previously been unable to perceive speech even with the most powerful hearing aids available. For a recent overview and discussion of the effects of auditory stimulation provided by hearing aids in comparison with cochlear implants in deaf children, see McCormick [33].

Following cochlear implantation, the effects of speech deprivation may be reversed, and this has been demonstrated for speech perception [34–41], and for speech production [42]. Staller et al. [34] investigated 80 children with 12 months experience of wearing a cochlear implant using a number of different speech perception tests. They found that significant negative correlations between the duration of deafness and performance on open-set measures of speech perception (spondee recognition, Central Institute for the Deaf sentences, and modified Glendonald Auditory Screening Procedure presented in open-set format). Hence, children with shorter durations of deafness were able to perform at higher levels than children with longer durations of deafness.

In a study of open- and closed-set speech perception, Dawson et al. [35] investigated four children whose age at implantation was less than 10 years, and whose duration of deafness was less than 8 years, and compared their results with four adolescents whose age at implantation was greater than 13 years, and whose duration of deafness was greater than 12 years. The effects of age at implantation and duration of deafness on open-set speech perception using the BKB sentence lists were marked. Of the four adolescents, two scored 0 and 2%, respectively, and the other two could not complete the task. In contrast, two of the three children who were tested were able to perform the task and received scores of 26 and 34%.

Fryauf-Bertschy et al. [36] investigated speech perception in ten congenitally deaf and three postlingually deafened children over 24 months of

implant use. They showed that postlingually deafened children performed best on word recognition using the Monosyllable-Trochee-Spoondee Pattern Perception Test, and that their rate of improvement was faster than for congenitally deaf children, with the improvement occurring within the first 6 months following implantation. These data further showed that congenitally deaf children implanted under 5 years of age performed better than congenitally deaf children implanted between 6 and 16 years of age. However, the authors were careful not to make this conclusion due to the preliminary nature of the study, and the small number of children involved.

Waltzman et al. [37] compared the performance of 14 children implanted between the ages of 2 and 3 years with 11 children implanted between 3 and 5 years of age. They found that speech perception performance at 2 years post-implantation was virtually identical for the two groups, with one exception being the Phonetically Balanced Kindergarten (PBK) word lists. This is an open-set test and so is much more difficult. Fewer children were able to complete the task. Nevertheless, Waltzman et al. found that children implanted between the ages of 2 and 3 years scored a mean of 62%, whereas the children who were implanted at a later age had a mean score of 22%. This difference was significant.

Kirk [38] has studied the effect of age at implantation in 56 prelingually deafened children who were either implanted early (2–5 years) or late (6–9 years). She controlled for the effect of experience by matching both groups for years of cochlear implant use. She used a test of lexical access and the PBK word list, and found a significant effect of age at implantation. Children who were implanted early performed better than those implanted late. This effect was apparent even though the children in the early group were around 3 years younger than children in the late group. Similarly, Clark ([39], Fig. 1) has reported speech perception results based on 44 children and young adults based on the BKB sentence lists, and found some indication for a sensitive period for speech perception up to approximately 4–6 years of age.

Tyler et al. [40] reported on the performance of 50 prelingually deaf children with a minimum of 2 years experience with cochlear implants measured on a variety of speech perception tests. Their data (their figure 4) collected using the PBK word lists on 13 children aged from 2 to 4 years of age at implantation, and on 15 children aged from 5 to 12 years of age at implantation shows that the children who were implanted in the earlier age group performed at a higher level at 2 and at 3 years following surgery.

Recently, Waltzman and Cohen [41] have studied open-set speech perception in nine profoundly deaf children who were provided with a cochlear implant when aged from 14 to 23 months. They retrospectively compared the performance of nine children implanted under the age of 2 years with that of 38 children implanted between 2 and 5 years of age on the Glendonald Auditory Screening Procedure word and sentence tests, and the PBK word lists. They found that the children implanted under the age of 2 years achieved similar levels of speech perception performance to their older counterparts. On the Glendonald sentence test, the younger children often outperformed the older group.

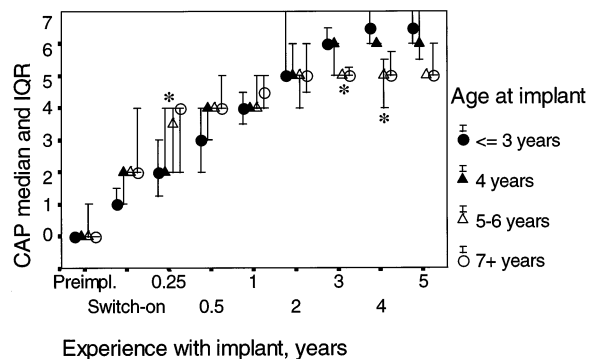


Fig. 1. Median and inter-quartile ranges for the Category of Auditory Performance (CAP, Archbold et al. [42]) for 62 deaf children with cochlear implants as a function of experience with the implant in years. The parameter is age at implantation, with four groups of children where surgery occurred in the third year of age or under, fourth year of age, fifth to sixth years of age, and seventh year of age onwards. Asterisks indicate significant differences between the groups using the Median test ($P < 0.05$).

Data from our own group provide further information on the effect of age at implantation. These data are derived from a rating scale of auditory performance devised by Archbold et al. [43] which is completed by teachers of the deaf. These data reveal the differential effects of age at implantation. Because children with a late age at implantation are older, they are more likely to perform better in the first few months post-implantation. In contrast, children with an early age of implantation are younger, and so initially do not perform as well in the first few months post-implantation. However, at 3 years following surgery, they perform better than children implanted at an older age. Fig. 1 shows the improved rating of auditory performance following experience of a cochlear implant in 62 children who were followed up for 3 years. The children were divided into four groups based upon age at implantation (less than or equal to 3 years, 4 years, 5–6 years, and 7 years and over). The numbers of children in each group are presented in the legend in Fig. 1. At the time of switch-on, the older children who were implanted late (7 years and over) perform better than children implanted at an earlier age. At 1 year following surgery, all groups have similar ratings, and by 3 years following surgery, children who were implanted at ages up to 4 years have higher ratings than those who were implanted at a later age. It is emphasized that these data are preliminary, and further analysis is underway [44]. Nevertheless, the data are consistent with earlier results [34–41].

Turning to the effects of age at implantation on speech production, Tye-Murray et al. [42] reported on 28 prelinguistic children with 3 years of implant experience. Children were divided in a group implanted at 2–4, 5–8 and 8–15 years. Tye-Murray used a measure based on phonetic transcriptions for phonemes and for words. They found that children who were implanted at under 5 years of age showed better performance than children implanted at older ages.

2.3. Multiple sensitive periods associated with language acquisition

So far, the studies reviewed have considered language development as a single unitary stage.

However, it is likely that language acquisition is not a single sensitive or critical period as originally conceived by Lenneberg [24]. Rather, there are several sensitive periods associated with various aspects of language [45,47,48]. Ruben [45] has recently argued for a number of sensitive periods of language acquisition, and has suggested that phonological development occurs by the end of the first year of age, followed by the development of semantics up to 4 years of age, and syntactic development up to 15–16 years of age.

Neville [13] reviews a number of studies that show that in normal hearing children of 4 years of age, an adult-like semantic processing response was elicited. The syntactic processing response does not mature until 15–16 years of age. The experimental basis for these insights is the now classic study of Neville et al. [46], who showed different electrophysiological patterns in the processing of English sentences in hearing adults. They found that an event related potential in posterior regions in both hemispheres (N400) associated with semantic processing (open class words, such as nouns, verbs and adjectives referring to specific objects and events), and another discrete potential (N280) in anterior regions of the left hemisphere associated with syntactic processing (closed class words including articles, conjunctions, and auxiliaries that specify relations between objects and events).

Weber-Fox and Neville [47] capitalized on the results found by Neville et al. [46] in a study of 61 adult Chinese/English bilinguals, who were exposed to English at different points in development: 1–3, 4–6, 7–10, 11–13, and after 16 years of age. Few differences were observed in the semantic processing response up to 10 years of age. In comparison, large response differences were found in the syntactic processing response, which indicated a longer period of maturation. A further example is the recent study of Neville et al. [48] who recorded event-related potentials from native deaf and hearing signers, and from hearing subjects who acquired sign late or not at all. Their results show that the response associated with semantic processing of sign language was identical to that found in hearing subjects. This response matured around 4 years of age of acquisition. The

syntactic processing response in the posterior area of left hemisphere found in hearing subjects was not found in deaf signers. Instead, a slightly earlier response around 250 ms was found at posterior sites in both hemispheres. Overall, the pattern of results found in this series of studies [13,32,47] suggest that the nature and timing of sensory and language experience affects the development of language systems of the brain. Furthermore, there are specialized subsystems that process different aspects of language processing, and these appear to have different sensitive periods.

An intriguing, and related finding has recently been announced by Reilly et al. [49], who have investigated morphology, syntax and narrative structure in children with focal lesions from 3 to 8 years of age on a longitudinal basis. In comparison with a group of normal control children, they found that the lesion group always performed worse, but recovered function with time. However, as children moved to another level of development in language acquisition, differences between the two groups of children recurred, and functional recovery was again observed in the lesion group again. It would be interesting to ascertain whether a similar or entirely different developmental pattern may be found in a group of implanted children. It is likely that longitudinal studies of language acquisition in profoundly deaf children with cochlear implants will provide further information on the inter-relationships between the sensitive periods.

2.4. Summary

Research on children with cochlear implants suggests that there are sensitive periods in which different aspects of language are acquired. Most, if not all, of the research cited in this second section of the review is preliminary, and so readers must be careful not to over-interpret the results. For example, it would be inappropriate to set up rigid clinical selection criteria based on age at cochlear implantation. There are large individual differences observed in all of the data reviewed, where some children who are older perform exceptionally well, and some children implanted under 5 years of age perform rather

poorly. Before such clinical selection criteria may be established, it is necessary to understand more fully why these large individual differences exist. Moreover, further data on the cascade of sensitive periods, and the study of their inter-relationship is important in the measurement of the different aspects of language acquisition (e.g. phonology, semantics, syntactic development).

3. Implications for future research

There appears to be a number of sensitive periods of language acquisition, rather than a single critical period as suggested by Lenneberg [24]. Moreover, the physiological data shows that learning and concomitant neural reorganization carries on into adulthood, albeit at a slower rate. Sensory activity leads to neural development, and the sustained effects of inactivity can lead to a loss of responsiveness in the auditory system. These effects may be reversed by the subsequent provision of sensory stimulation, such as that delivered by cochlear implants [15–21].

The study of language acquisition in children with cochlear implants provides us with information about the developmental plasticity of the auditory system. The evidence suggests that congenitally deafened children implanted under the age of around 5 years are likely to perform better on speech perception and speech production tasks than children implanted at an older age [36–41]. It would be useful to study individual differences to provide information on environmental influences that may adversely affect or improve performance with a cochlear implant.

There appear to have been no studies investigating the effect of a cochlear implant on the compensatory visual and somatosensory development that occurs in deaf children and adults [13,14,32,45,46]. Research in this area is likely to further reveal the importance of biological considerations for language acquisition [24].

Based on preliminary electrophysiological measurements, Ponton et al. [15] have suggested that there is a maturational delay in implanted children that approximates the period of auditory deprivation prior to implantation. This may indi-

cate that the auditory system has some capacity to resume the normal developmental sequence after a period of auditory deprivation. Further studies relating the electrophysiological findings with measures of language development are required to assess the clinical significance of this result. In addition, there is likely to be an interaction with compensatory plasticity which may or may not have limited the resumption of a normal auditory developmental sequence.

Finally, Ruben [43] has suggested that there are a number of sensitive periods that characterize language acquisition, and has called for detailed studies of phonological, semantic and syntactic development. It is likely that longitudinal studies on the language acquisition of profoundly deaf children with cochlear implants will enable us to better understand this complex area.

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