Letter to the Editor: Do Pediatric Cochlear Implant Recipients Display Domain-General Sequencing Difficulties? A Comment on Davidson et al. (2019)

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A recent study by Davidson et al. (2019) addressed an issue that is of considerable importance to the field of cognitive hearing sciences: to what extent does hearing loss result in changes to cognitive functions outside of hearing per se? From a traditional audiological perspective, hearing loss might be thought to primarily affect hearing, audibility, speech perception, and spoken language. However, because the auditory cortex is connected to other regions of the brain that carry out domain-general processing, it should not be overly surprising if hearing loss, especially hearing loss early in life, results in changes to nonauditory functions (Kral et al. 2017). Empirical studies have not been conclusive on this matter, and exactly which cognitive functions aside from auditory perception are affected by hearing loss remains an open and important question.

Davidson et al. (2019) attempted to address this question by examining whether working memory deficits in deaf children with cochlear implants (CIs) were domain specific or domain general. Working memory tasks were created that differed in terms of task difficulty (simple versus complex) and processing domain (verbal versus visuospatial). The simple working memory tasks required participants to recall and reproduce sequences of stimuli; the complex tasks required participants to perform an additional secondary task while engaging in the primary recall task. The verbal working memory tasks employed stimuli that were easy to encode and rehearse using verbal strategies (i.e., sequences of digits: "2"-"7"-"3," etc.), whereas the visuospatial tasks used stimuli that afforded no obvious way to verbally code them (i.e., pointing to and clicking a sequence of locations displayed on the screen). Two groups of children were tested on these novel tasks. One group consisted of 25 deaf children 5 to 9 years old who had at least one CI, had received educational services via spoken language, and had no reported delays in cognitive development. It should be pointed out that the sample of deaf children was relatively heterogeneous in terms of whether the children had bilateral CIs or bimodal devices as well as in terms of the etiology of hearing loss. A second group consisted of 29 age-matched children with normal hearing (NH) who also had no reported delays in cognitive development.

To analyze the data, Davidson et al. (2019) converted the raw scores on each task to z scores based on the distribution of data from the CI and NH groups combined. The authors then conducted an analysis of covariance (ANCOVA) on the z scores with age at test and maternal education entered as covariates, group (NH versus CI) as a between-subjects variable, and domain (verbal versus visuospatial) and task difficulty (simple versus complex) as within-subject variables. The results revealed a main effect for group and no interactions, with the NH group performing better on all four versions of the working memory tasks relative to the CI group. Davidson et al. (2019) next examined the 95% confidence intervals for the CI and NH groups on each task (their Figure 2). The authors observed that the verbal tasks showed a clear separation in the distributions between the NH and CI groups, whereas there was some amount of overlap between the groups for the visuospatial tasks. The authors interpreted these data as showing that "CI users exhibited significantly lower performance relative to NH peers on tasks that required some degree of verbal processing … and exhibited similar performance on visuospatial processing tasks" (p.7). Davidson et al. (2019) concluded that "children with CIs have domain-specific deficits related to storing and processing verbal information in working memory" (p.10) and that "their ability to store and process visuospatial information … seems to be intact and on par with their NH age mates" (p.10). Davidson et al. (2019) suggested that these "domain-specific effects" are due to children with CIs receiving impoverished auditory input, which disrupts their efficient and accurate encoding of verbal information, leading to deficits in verbal working memory but not visuospatial processing.

A CLOSER EXAMINATION

Davidson et al. (2019) should be commended for their overall approach and their use of elegant tasks that provide intriguing data into the nature of information processing in children with CIs. However, we believe that their interpretation of the data is incomplete and that additional inspection of their results reveals potentially important differences in cognitive processing exhibited between the NH children and pediatric CI recipients. For example, although the authors interpreted the slightly overlapping confidence intervals for the visuospatial tasks as evidence of no group differences, the presence of overlapping confidence intervals alone does not confirm a lack of statistically significant differences between groups. Specifically, a lack of overlap in confidence intervals indicates significant group differences (Cumming & Finch 2005), but the opposite conclusion, that overlapping confidence intervals necessarily imply lack of group differences, is not true (Knol et al. 2011). In fact, as a "rule of eye" for comparing means between two independent groups, 95% confidence intervals can overlap as much as 50% and still be significantly different at $p < 0.05$ (Cumming & Finch 2005). An inspection of Figure 2 in Davidson et al. (2019) reveals that the amount of overlap of the

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0196/0202/2019/XXXX-00/0 • Ear & Hearing •

<zdoi; 10.1097/AUD.0000000000000841>

confidence intervals between the two groups for the complex visuospatial task may be perhaps >50%, but the overlap for the simple visuospatial task appears to be <50%. Thus, based on an examination of the confidence intervals alone, and without additional analyses, a significant group difference appears to be possible for at least one of the visuospatial tasks in addition to the two verbal working memory tasks.

Examining the effect sizes for the differences between groups on these four tasks can provide additional information about potential group differences. Although Davidson et al. (2019) did not report effect sizes for these comparisons, Cohen *d* effect sizes can be calculated based on the means, SDs, and sample sizes provided in their Table 2. The results of these new calculations indicated medium- to large-sized differences between the NH and CI groups for both verbal tasks (d = 0.75 and 0.6 for the simple verbal and complex verbal tasks, respectively) and small- to medium-sized differences between the groups for both visuospatial tasks (d = 0.25 and 0.29 for the simple and complex visuospatial tasks, respectively; Cohen 1992). Note that these values are based on calculations involving the raw scores, but similar effect sizes should be obtained when using the *z* scores instead. Although the effects are larger for the verbal tasks relative to the visuospatial tasks, effect sizes in the small-to-medium range are not negligible (Meyer et al. 2001) and would appear to suggest that as a group, the deaf children with CIs may have had lower span scores than the NH children on the visuospatial as well as the verbal tasks.

Perhaps most importantly, the omnibus ANCOVA showed no significant interactions and only a main effect of group, which does not support a conclusion of different effects for the different types of memory tasks between groups. Although the lack of significant interaction effects may be a result of insufficient power as opposed to a true absence of differences between verbal versus visuospatial memory tasks, caution in drawing conclusions about differences between these tasks appears to be warranted. Just as one should not accept the null hypothesis and conclude statistical equivalence based on a failure to find a statistically significant difference (without conducting a formal equivalence analysis, Lakens 2017), one can also not assume that the difference between a statistically significant result and a nonstatistically significant result is, on its own, significant. That is, "the difference between 'significant' and 'not significant' is not itself statistically significant" (Gelman & Stern 2006). Certainly, the article by Davidson et al. (2019) is not the only one to contain such errors in statistical interpretation; however, it is the focus of this letter because it relates to an important question and debate in the field about the nature of the consequences of hearing loss.

In summary, the lack of significant ANCOVA interaction effects, combined with implications of overlapping confidence intervals and our analysis of effect sizes, supports a more nuanced interpretation of the study's results. We thus believe the most appropriate conclusion is that children with CIs exhibited lower performance across all four working memory tasks, regardless of domain or task complexity. It appears likely that the effects are larger for the verbal tasks, but there appear to be group differences for the visuospatial tasks as well; a study powered to detect small-to-medium effect sizes would be needed to fully test this pattern of differences between groups.

UNDERSTANDING DOMAIN-GENERAL EFFECTS OF HEARING LOSS

Why might poorer performance for pediatric CI users be observed on nonverbal, visuospatial working memory tasks? Clearly, hearing loss results in difficulties with hearing, speech, and language processes that rely upon manipulation of phonological or verbal input (as recognized and discussed by Davidson et al. 2019), which explains the group differences on the two verbal working memory tasks. However, there is also a body of evidence suggesting that differences in auditory perception and spoken language processes are not the only effects of hearing loss (Bharadwaj et al. 2012; Bharadwaj & Mehta 2016; Deocampo et al. 2018; Pisoni & Cleary 2004; Pisoni et al. 2016; UIanet et al. 2014). If one recognizes that the brain is a massively interconnected complex system, then it should not be surprising that a loss or disturbance in one sense may affect the functioning and integrity of other neural and cognitive systems, especially if the sensory loss occurs early in development (Luria 1973; Myklebust & Brutten 1953). For instance, children who are deaf and use American Sign Language (without CIs) show different patterns of attention to visual stimuli compared with typical hearing children, with attention being more distributed and focused toward the visual periphery (e.g., Dye & Bavelier 2010; Dye et al. 2007). If central processes of (nonauditory) attention are affected by the presence of hearing loss, then it is possible that visuospatial working memory could also be affected, given that working memory and attention are closely linked and heavily intertwined (Awh et al. 2006).

We have previously proposed that hearing is intimately connected to cognitive processes related to the encoding, storage, and retrieval of serial order information (Conway et al. 2009; Hirsh 1967). Research has shown that for information processing tasks that require encoding events or stimuli in serial order, people generally perform better when hearing can be relied upon, rather than vision, even for nonlinguistic materials (Conway & Christiansen 2009). This hearing-sequencing connection is due to the fact that auditory signals are inherently temporal and sequential in nature, whereas for vision, serial order is not obligatory in the same way. Thus, sound may play an important organizing role in shaping cognitive development by providing important early experience—and experience-dependent learning—with serial and temporal information, which supports and "scaffolds" the development of information processing skills related to sequence processing, learning and memory, and serial order behavior more generally. One consequence of this "auditory scaffolding hypothesis" (Conway et al. 2009) is that hearing loss early in perceptual and cognitive development reduces the amount of experience with sequential input, due to lack of exposure to sound, which results in atypical development of and disturbances in serial order information processing skills, even for visual and nonverbal sequential input. Note that in this formulation, it is auditory experience specifically, not language experience (Hall et al. 2018), that promotes the development of sequence processing skills. Although spoken language is an excellent example of an auditory sequential signal (cf., Poeck & Huber 1977), other auditory signals such as music also convey rich sequential structure (Patel et al. 1998). Thus, lack of exposure to sound implies less exposure with such heavily sequenced input, which we argue can affect the development of skills related to serial order processing.

The phenomenon of “diaschisis” provides a potential explanatory neural mechanism for the postulated consequences of auditory deprivation. Diaschisis occurs when abnormalities with or atypical development of a particular brain region (such as auditory cortex due to auditory deprivation) results in loss of input and neural firing to “downstream” brain regions, resulting in changes and potential weakening of the structural and functional connectivity of those regions (Carrera & Tononi 2014). One crucial brain region that lies downstream from auditory areas is the prefrontal cortex (Kral et al. 2016), which is known to be critically important for mediating sequence processing and serial order behavior (Fuster 2001). It is interesting that some data indicate that individuals who are deaf show atypical functioning of frontal areas of the brain (Wolff & Thatcher 1990).

Thus, the mechanism of diaschisis can potentially explain why performance on nonauditory and nonverbal visual-spatial sequence processing tasks is atypical in some individuals who are deaf or hard of hearing. Hearing loss changes the structural and functional integrity of the auditory cortex (e.g., Xia et al. 2017), which in turn could lead to diaschisis-mediated cortical reorganization in brain areas such as the prefrontal cortex, affecting sequence processing as well as other cognitive skills such as working memory, cognitive control, and executive function (Kral et al. 2016). In addition to the recent findings reported by Davidson et al. (2019), a number of other recent studies have also found atypical sequence processing in nonauditory and nonlanguage domains in individuals who are deaf or hard of hearing (Bharadwaj et al. 2012; Bharadwaj & Mehta 2016; Gremp et al. 2019; Lévesque et al. 2014; UIanet et al. 2014; though for conflicting evidence, also see Giustolisi & Emmorey 2018; Hall et al. 2018; Torkildsen et al. 2018).

However, the auditory scaffolding hypothesis does not preclude there also being domain-specific effects resulting from hearing loss, in addition to the more global and domain-general changes discussed above. That is, hearing loss can directly affect spoken language development, which in turn creates difficulties for cognitive processes that rely upon or are facilitated by the use of verbal labeling or active verbal rehearsal processes (Davidson et al. 2019). But in addition to this domain-specific effect, with properly sensitive paradigms such as the one used by Davidson et al. (2019), nonauditory and nonverbal effects may also be observed. Such a combination of domain-specific and domain-general consequences of hearing loss was also observed in a recent study that attempted to control for verbal labeling and rehearsal by using visual sequence learning and memory tasks with stimuli that were either easily nameable (sequences of colors) or more difficult to name (sequences of noncolored visual-spatial locations) (Gremp et al. 2019). The results of this study, like those from Davidson et al. (2019), indicated that children who were deaf or hard of hearing displayed lower levels of sequence learning and memory for both types of stimuli, but showed the biggest differences for the color sequences that were comparatively easier to verbally label.

In addition, the auditory scaffolding hypothesis does not preclude effects of other influences on domain-general neurocognitive processes, which may mitigate or exacerbate the effect size of hearing loss. Multiple biopsychosocial factors, including hearing loss, interact in a complex, systemic process to influence neurocognitive development. Environmental experiences (including enrichment and family environment), education, and clinical interventions are only a few of the compensatory factors that may interact with hearing loss in influencing neurocognitive outcomes. As a result, large individual differences in sequence processing and other domain-general neurocognitive outcomes are routinely found in samples of children with hearing loss (Kronenberger & Pisoni, Reference Note 1).

Thus, taken together, the recent data from Davidson et al. (2019) suggest that hearing loss leads to difficulties and disturbances in processing verbal input, but difficulties with broader, domain-general sequencing abilities cannot be ruled out. Additional work is needed to confirm the nature of domain-general sequencing abilities in children who are deaf or hard of hearing. It could be fruitful to examine performance for children who vary on the duration of auditory deprivation, age of first device use, and severity of hearing loss as a way to tease apart which factors are associated with any such processing difficulties.

CONCLUSION

The question is not whether hearing loss affects domain-general cognitive functions, but what functions are affected and how they are changed. This information in turn is vital for informing intervention approaches. The auditory scaffolding hypothesis proposes that one such affected domain is sequential learning and processing, mediated by changes to anterior brain networks such as the prefrontal cortex as a result of diaschisis (Conway et al. 2009; Conway et al. 2011). A number of research findings, including those from the recent study by Davidson et al. (2019), appear to be consistent with the core hypothesis that sequence processing difficulties persist in individuals with hearing loss, even for nonlanguage and nonauditory information processing domains (Bharadwaj et al. 2012; Bharadwaj & Mehta 2016; Gremp et al. 2019; Lévesque et al. 2014; UIanet et al. 2014). There are likely other domain-general functions that are also affected by hearing loss, such as visual attention (Dye & Bavelier 2010; Dye et al. 2007) and inhibitory control processes (Kronenberger et al. 2013; Kronenberger et al. 2014). Additional research is needed to better specify which functions are affected and how. A better understanding of these effects holds the promise for formulating new, more robust interventions and techniques for improving overall cognitive and language function in individuals with hearing loss who are at risk for suboptimal outcomes.

ACKNOWLEDGMENTS

C.M.C. wrote the main paper. All authors discussed and commented on the manuscript at all stages. We would like to thank Angela AuBuchon, Adam Bosen, Monita Chatterjee, Samantha Emerson, Katherine Gordon, and Kaylah Lalonde for their helpful comments on earlier versions of this manuscript.

This work was supported in part by the National Institute on Deafness and Other Communication Disorders: R01DC012037 (C.M.C.) and R01DC015257 (D.B.P., W.G.K.).

W.G.K. is a paid consultant to Shire Pharmaceuticals, Homology Medicines, and the Indiana Hemophilia and Thrombosis Center (none are relevant to the current article). The other authors have no conflicts of interest to disclose.

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