Combination of Comorbidities as a Reference Standard for Evaluating Auditory Processing Disorder: Response to the Letter to the Editor From Schow et al. (2021)

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Purpose: Concerns expressed by Schow et al. (2021) around the evidence-based diagnostic criteria suggested by Ahmmed (2021a) are addressed here. The use of combination of comorbidities as a reference standard for evaluating auditory processing tests is a valid strategy, consistent with the Research Domain Criteria framework from the National Institute of Mental Health as well as the Standards for Reporting of Diagnostic Accuracy Studies. The correlations between auditory processing tests and some comorbidities in Ahmmed (2021a) were significant at $p < .01$. The low sensitivity and specificity reported was not related to the principle of using comorbidities as a reference standard but due to the choice of comorbidities combined in the reference standard. Ahmmed (2021a) suggested the option of inclusion of other comorbidities in addition to language impairment and impaired manual dexterity in the reference standard. Visual processing impairment could be considered as the additional comorbidity to improve sensitivity and specificity of the approach suggested by Ahmmed (2021a).

We thank Schow, Dillon, and Seikel (2021) for their feedback to our article and the American Journal of Audiology for the opportunity to respond. The ongoing impasse surrounding the conceptual and diagnostic basis of auditory processing disorder (APD) is well known (Moore, 2018). We hope that the Ahmmed (2021a) article, the letter by Schow et al. (2021), and this response will stimulate further discussions to improve our understanding of APD and developing new strategies.

Issue 1: Advance the Goal of Evidence-Based Diagnostic Criteria for APD

Defining and rationalizing test positivity cutoffs is important for diagnostic accuracy studies (Cohen et al., 2016, Item 12, p. 7). The rationale of the American Speech-Language-Hearing Association (ASHA, 2005, p. 13) criteria of 2 SD below the mean (2nd percentile) in two auditory processing (AP) tests for diagnosing developmental APD is unclear and fail to diagnose APD in a significant number of children with functional difficulties (Dawes et al., 2008). The first aim of the Ahmmed (2021a) study was to explore the appropriateness of the ASHA (2005) criteria when co-morbid neurodevelopmental conditions (NDCs) are diagnosed using the 5th percentile cutoff or higher—for example, attention-deficit/hyperactivity disorder (ADHD; Swanson et al., 2012) and developmental coordination disorder (DCD; Smits-Engelsman et al., 2015). The combination of AP tests and tools for comorbidities was considered at a second stage, from ASHA (2005, p. 10) considering significant functional difficulties and DeBonis (2015) considering failing one of two questionnaires (CCC2 [Bishop, 2003] and BRIEF [Gioia et al., 2000]), for diagnosing APD following only one AP test fail. Schow et al. (2020) also used combination of APD tests plus a questionnaire but did not report the evidence for the cutoff they used. It also needs to be noted that all the tools assessing comorbidities in Ahmmed (2021a) were not questionnaires.

Disclosure: The author has declared that no competing financial or nonfinancial interests existed at the time of publication.
Issue 2: Meeting the Core Standards for Reporting of Diagnostic Accuracy Studies (STARD) Components

In accordance with the STARD guidelines (Cohen et al., 2016), developmental APD (with or without comorbidities) was the “target condition,” with the five SCAN-3 tests (Keith, 2009a, 2009b) as “index tests,” and the presence of one or more of three comorbidities as the “reference standard.” Inclusion of children with “suspected APD” due to presence of different functional symptoms is consistent with the STARD (Cohen et al., 2016, Item 6, p. 5) “eligibility criteria.”

Issue 3: Appropriateness of Using Comorbidities as the Reference Standard

A reference standard needs to be a reliable and valid measure to define the target condition (Cohen et al., 2016). There are concerns that reference standards used for most current tests evaluating developmental APD do not fulfill these requirements (Vermiglio, 2018). Schow et al. (2020) defines their reference standard as APD identified formally by local professionals using different methods and techniques. In this instance, the reference standards and the norms of tests used by the different professionals lacked clarity, and the reliability and validity of such an approach is questionable. Schow et al. (2020, p. 1001) also considered the rationale of using comorbidity as a reference standard. The authors themselves expressed concerns that their method was not ideal. Schow et al. (2020) wondered what an ideal reference standard would be and made an interesting comment that any method would lead to objection. Our opinion is that the use of appropriate combination of comorbidities using very definite cutoff criteria is a concrete, transparent, and replicable reference standard consistent with the STARD 2015 guidelines compared to Schow et al. (2020).

The high degree of comorbidity between developmental APD and related NDCs is almost universally accepted (de Wit et al., 2018; Moore, 2018; Schow et al., 2020, pp. 995, 1002), with APD being considered as a marker of a neurodevelopmental syndrome (Moore & Hunter, 2013) or spectrum disorder (Wilson, 2019, p. 246). Due to the heterogeneity and overlap, the National Institute of Mental Health (NIMH, 2019) suggested a Research Domain Criteria (RDoC) framework and highlighted the importance of considering such multiple NDCs together in diagnostic formulation to capture the complex impact on individuals (Harrison et al., 2019). Therefore, the use of a combination of NDCs as a reference standard is consistent not only with ASHA (2005, p. 8) but also with the more recently developed RDoC framework (NIMH, 2019).

Schow et al. (2020) highlighted the importance of APD evaluation in children with NDCs. In a similar vein, it is important that children with APD are also evaluated for the possibility of undiagnosed NDCs. This is consistent with the International Classification of Functioning, Disability and Health (ICF) of the World Health Organization (WHO, 2001/2017) principle to address “functioning at the level of the whole person.” NDCs such as ADHD, high-functioning autism spectrum disorder (ASD), DCD, and language disorders, among others, are well established in neurodevelopmental pediatrics with widely available assessment and management pathways. Diagnosis of comorbid NDCs takes diagnostic precedence over APD (British Society of Audiology, 2018), and such an approach not only allows using comorbidities as a reference standard but also ensures targeted support for both APD and other NDCs that were not suspected earlier (Alonso Soriano et al., 2015; Wilson, 2019).

Ahmmed (2021a) used AP tests evaluating three auditory domains (dichotic listening, monaural low-redundancy speech perception, and temporal processing), similar to Schow et al. (2020). This would allow identification of different combinations of auditory-perceptual deficits, irrespective of comorbidities present. Therefore, if APD is used as an umbrella term to refer to listening difficulties in the presence of normal pure-tone audiogram, the use of comorbidities as a reference standard should not interfere with the distinctiveness of developmental APD.

Issue 4: Comorbidity Approach Increasing the Diagnostic Rate of Both Comorbidity Conditions and APD

The APD tests are used to identify listening difficulties in different domains, and these APD tests will not identify NDCs. Similarly, validated tests with definite cutoff scores widely used in routine neurodevelopmental pediatrics to identify different NDCs are not going to diagnose APD. The fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013) and the RDoC framework (NIMH, 2019) allow identification of these separate conditions on their own right, even when they overlap. Therefore, the practice of holistic evaluation of children with suspected APD outlined in Ahmmed (2021a) is not going to overdiagnose any of the comorbid conditions. The data presented in Ahmmed (2021a) show four different outcomes: one without APD or comorbidities, one with APD only (pure APD, without comorbidities), one with both APD and comorbidities present, and another with only comorbidities (without APD).

Issue 5: Relationship Between Variables

The high correlation coefficient of .703 between Competing Words-Directed Ear (CW-DE) and Competing Sentences (CS) can be explained by the fact that they both represent dichotic listening. In the Ahmmed (2021a) article, the correlation coefficients between CW-DE and CS with General Communication Composite (GCC), a measure of language impairment, were admitted below .703 but were still significant at $p < .01$. Moore et al. (2010) also reported significant correlations of GCC with frequency discrimination (coefficient $-19, p < .001$) and frequency resolution
(coefficient .10, \( p < .01 \)) tests. In the Moore et al. (2010) study, the high levels of significance with low correlation coefficients were due to the large sample size. In Ahmmed (2021a), the correlation coefficients between Manual Dexterity (MD) and CW-DE (coefficient .265), CS (coefficient .319), Time Compressed Sentences (TCS; coefficient .255), and Auditory Figure Ground 0 dB (APG0; coefficient .204) were all significant at \( p < .01 \) level. Significance at \( p < .01 \) is not considered to be erroneous (Cameron et al., 2016).

The clinical significance of the relationship between APD and impaired manual dexterity reported in Ahmmed (2021a) is consistent with the notion that motor development plays a foundational role in the development of cognitive, social, and perceptual skills (Libertus & Hauf, 2017). Transmodal impairments affecting listening, as well as bimanual motor skills, have also been reported by Bellis and Ferre (1999). Additionally, in recognition of the importance of motor impairment, a new domain of “sensorimotor system” was recently added to the RDoC framework (NIMH, 2019). Ahmmed (2021a) findings contribute to the APD literature by demonstrating that severity of APD is related to impaired motor skills.

The clinical significance of the relationship between auditory perception and language in Ahmmed (2021a) is consistent with link between auditory and linguistic processing recognized in the ASHA Practice Portal (ASHA, n.d.). The only comorbidity that failed to show any correlation with the AP tests was ADHD. However, ADHD was found to relate to language impairment, and this was consistent with the literature (Baird et al., 2000).

**Issue 6: Poor Sensitivity and Specificity**

The issue of the low sensitivity and specificity raised by Schow et al. (2021) has been discussed in the Ahmmed (2021a) article. We disagree with Schow et al. (2021) that it is due to the concept of using comorbidities as the reference standard. Our opinion is that the poor sensitivity and specificity of the Ahmmed (2021a) diagnostic system was the choice of the comorbidities included in the reference standard. Ahmmed (2021a) used the presence of one or more of ADHD, language impairment, and impaired manual dexterity as the reference standard, of which ADHD did not correlate with APD tests. There are number of other comorbidities that were not included, such as visual perception, which is one of the subconstructs along with auditory perception under the cognitive system of the RDoC framework (NIMH, 2019). Further studies are required to find the most appropriate combinations of comorbidities as a reference standard to explore the diagnostic efficacy of APD tests. Our department is currently analyzing data of children who were assessed for both APD and visual processing. Preliminary indications are that inclusion of visual processing impairment as one of the comorbidities will improve the sensitivity and the specificity, possibly better than the diagnostic system suggested in Schow et al. (2020). The findings will be presented for review for publication in due course.

The Ahmmed (2021a) article highlighted the discrepancy in clinical practice regarding the cutoff criteria for APD and comorbidities. Table 4 in Ahmmed (2021a) is consistent with the literature that the 2nd percentile criterion (ASHA, 2005) will miss significant numbers of children with symptoms of APD (Dawes et al., 2008), which will have impact on the quality of life of these children. It is incorrect for Schow et al. (2021) to say that Ahmmed (2021a) strategy will miss the pure APD cases, as they are represented in Table 4 as APD only.

A second phase of tests exploring AP domains such as pattern recognition, gap detection, spatial processing, among others, is indicated in the small proportion of children who pass the three AP domains in the Ahmmed (2021a) study. This will identify additional children with APD (Ahmmed, 2021b).

**Issue 7: Severity and Nature of APD**

The comments by Schow et al. (2021) imply that the authors accept the 2nd, 5th, and 9th percentile criteria used in Ahmmed (2021a) as different levels of severity of APD. But, if one follows the ASHA (2005) 2nd percentile cutoff to diagnose APD, the children identified to have APD using the 5th and 9th percentile cutoff by Ahmmed (2021a) would not be diagnosed as APD. Therefore, a consensus is needed to define and rationalize the cutoff required to diagnose APD (Cohen et al., 2016, Item 12, p. 7).

The RDoC framework (NIMH, 2019) suggests a number of domains to understand the nature of co-existing conditions, and these domains include language, auditory perception, and motor skills, aspects that were reported in Ahmmed (2021a). Therefore, it is not correct that the Ahmmed (2021a) strategy would not help in understanding the nature of APD. We hope that this understanding will improve in future by considering other constructs, such as visual perception in relation to APD.

**References**


