

An Affront to Scientific Inquiry: A Response to David Moore's Editorial: Auditory Processing Disorder.

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It was with great concern that we read David Moore's editorial on auditory processing disorder (APD) (April 16, 2018 - Volume Publish Ahead of Print). Moore begins his editorial noting that in a "highly detailed critique" of a recent submission on early diagnosis of APD, two of three reviewers questioned whether an earlier diagnosis would be useful "because the status of APD is so controversial" or "difficult to verify." Moore uses these anonymous reviewers' comments to justify, based on a few selected papers, several of which are opinion papers rather than research or systematic reviews, that, effective immediately, "articles that either implicitly or explicitly assume APD as a single diagnostic characteristic of the auditory system likely will not be considered for publication." This Editorial Board policy was adopted without presenting research (and perspectives) that present compelling arguments that would refute such a policy. Moore states that this new *Ear and Hearing* policy is "endorsed by the whole Board;" however, the editorial is signed only by Moore. Moreover, as clinicians trained in diagnosis, we do not understand Moore's conflation of the disorder of auditory processing and a "diagnostic characteristic." According to the Diagnostic and Statistical Manual of Mental Disorders, DSM-5 (American Psychiatric Association, 2013), a disorder is defined as "a syndrome characterized by clinically significant disturbance in an individual's cognition, emotion regulation, or behavior that reflects a dysfunction in the psychological, biological, or developmental processes underlying mental functioning"(p. 20). Thus, by definition, a disorder is not a "diagnostic characteristic." We provide scientific evidence in this rebuttal that challenges a multitude of Moore's statements and conclusions used to support his opinion and this indefensible editorial policy. We note the considerable adverse consequences of this policy for scientific inquiry and research, dissemination of scientific knowledge, and ultimately good patient care, and we conclude by urging the Editorial Board to reverse this biased policy.

APD Is A Well-Established Disorder

We begin by emphasizing that the submission that caught Moore's attention and was (presumably) rejected was done so not as a result of methodological design flaws, but rather because the reviewers considered the disorder *controversial and difficult to verify*. This opinion stands in stark contrast with the inclusion of APD in both the International Classification of Disorders manual of the World Health Organization version 10 and 11 beta version (ICD-10 and ICD-11 Beta version), in which APD is recognized as a distinct diagnosis and disorder, as it is by many professional associations and societies throughout the world (e.g., American Academy of Audiology, 2010; American Speech-Language-Hearing Association, 2005; International Bureau for Audiophonologie 2007; Canadian Interorganizational Steering Group for Speech-Language Pathology and Audiology 2012; Auditory Processing Disorder: New Zealand Review 2014; Danish Medical Audiological Society, 2014; Deutsche Gesellschaft für Phoniatrie und Pädaudiologie (DGPP) 2015). Moore thus devalues the ICD and professional association position statements and guidelines and specifically dismisses those of the American Speech-Language-Hearing Association (ASHA 2005) and the American Academy of Audiology (2010). Instead, he cites three papers to argue that "most if not all cases of childhood APD" really reflect learning or language disorders. To support that assertion, Moore cites de Wit et al.'s (2018) systematic review of 13 studies of moderate quality comparing the performance of children with suspected or diagnosed APD and children diagnosed with a different developmental disorder. This paper concluded that children with various developmental disorders performed similar on the outcome of 85 of 102 (sub)tests, including auditory tests and questionnaires and tests of intelligence, attention, memory, language, and reading. Further, de Wit et al. concluded that "listening difficulties experienced by children are multifaceted and that there is substantial overlap between various developmental disorders." They quite astutely proposed "therefore, it is crucial that various professions work together and use a multidisciplinary approach not only in the assessment of children with listening complaints but also in the event of children who satisfy the diagnostic standards of SLI,

ADHD, and dyslexia” and, contrary to Moore’s editorial, concluded that “additional research is required to better understand the different profiles of children with various complaints or disorders”. If *Ear and Hearing* bans research on APD, presumably, neither review papers like de Wit et al.’s nor research papers like the ones de Wit et al. call for would be published. Obviously, these types of high quality research papers are needed to advance science not only in the field of APD, but also for developmental disorders in general.

Moore also cites DeBonis (2015) and Cameron et al. (2014) whose work actually support the APD diagnosis. While several of us published a rebuttal to DeBonis (2015) (Iliadou, Sirimanna, Bamjiou, 2016), DeBonis does note the adverse impact of speech recognition in noise deficits—well established as a characteristic processing disorder in APD (AAA 2010; ASHA 2005), and Cameron et al. focus on “spatial processing disorder” which involves the interplay of speech recognition in noise with two other well established auditory processes--- localization and binaural interaction. Rather than refuting the existence of APD, these reports substantiate the diagnosis. There are 326 peer reviewed APD papers in PubMed and over a thousand APD papers in the Scopus database: only a handful deny the existence of APD, and none of that handful provide conclusive evidence to support that assertion.

A recent European consensus on APD reached by professionals in 17 European countries (Iliadou et al. 2017) noted that the two primary reasons why debate around APD lingers are: (1) failure by critics to employ the currently available auditory processing test battery, with documented sensitivity and specificity, leading them to (2) draw conclusions regarding APD based on characteristics of individuals who are not diagnosed with APD and are so-called “*suspected APD*”. Interestingly, Moore ignores this multi-authored paper but selectively cites single-authored opinion papers, that are consistent with his perspective, rather than cite this European consensus paper or other opinion and research papers that counter his viewpoint (e.g., Chermak et al. 2017; Halliday et al. 2017; Iliadou et al. 2017; Iliadou and Kiese-Himmel 2018; Abramson et al. 2018), and other research articles presenting the current mainstream clinical diagnostic and treatment approach (Brenneman et

al. 2018; Musiek et al. 2018). Moore references a single book focused on APD controversies, but fails to cite the numerous books that focus on the auditory neuroscience underlying, and the practice of, diagnosing and treating APD (e.g. Bellis, 1996; Chermak and Musiek 2014; Musiek and Chermak 2014; Rawool 2016; Geffner and Ross-Swain 2019).

Few if any professionals active in the field of audiology would deny the potential for cognitive and linguistic confounds when evaluating auditory processing or any task that requires cooperation and behavioral response on the part of the individual being tested, not least because the client with any developmental disorder is at a higher risk of having more than one disorder than the general population (e.g. in the case of ADHD Attention deficit hyperactivity disorder: diagnosis and management, NICE guideline, September 2017 or autism (Mody and Belliveau 2013). There are, however, several tutorials on how to best minimize cognitive and linguistic confounds in diagnosing APD (e.g., Chermak et al. 2017) with a goal to more fully inform individualized treatment and management for clients with complex and diverse needs.

APD is a Disorder of the Central Auditory Nervous System

Moore selectively recounts the history of APD, citing the observation by Myklebust (1954) of children with normal hearing sensitivity with listening difficulties. This observation should be viewed, however, within the historical context of work that began in Italy by a team of otorhinolaryngologists, led by Ettore Bocca, which confirmed that patients with central auditory nervous system (CANS) lesions presented significant deficits processing degraded speech, despite the presence of normal audiograms (Bocca et al. 1954). Bocca et al. provided indisputable evidence for the neurological origins of APD and established the cornerstone upon which APD is diagnosed today. The neurological subtypes of APD have since been extensively documented (see for example Griffiths et al. 2012; Bamiou et al. 2012; Grube et al. 2016; Koohi et al. 2017; Murphy et al. 2017; Iliadou et al. 2017; Polanski et al. 2017; Harris and Dubno 2017; Szymaszek et al. 2017; Iliadou and Eleftheriadis

2017; Durante et al. 2018). Moreover, Boscaroli and colleagues have demonstrated using neuroimaging that children with perisylvian polymicrogyria, which are indicative of functional cortical abnormalities, present with APD. Boscaroli and colleagues documented a correlation between degree of auditory processing deficits and extent of cortical abnormality. The performance of children with perisylvian polymicrogyria on central auditory processing tests was, indeed, similar to the performance of children with ‘developmental’ CAPD (i.e., children with no hard evidence of CANS lesions) on these same central auditory processing measures (Boscaroli et al. 2010, 2011; Amaral et al. 2015). Boscaroli’s work documents an anatomical correlate (polymicrogyria) to central auditory test findings. Further evidence for a neurological underpinning of APD comes from the long course of myelin maturation that has been shown in children to correlate with their ability to perform on tests of dichotic listening (i.e., revealed as left ear deficits). The left ear deficit is a consistent and dependable measure reflecting interhemispheric transfer of acoustic information (Berlin et al. 1973; Musiek et al. 1984; Boles et al. 2008; Bellis et al. 2008; Westerhausen, Bless, Kompus, 2015).

The Test Battery Gold Standard

In a section, that Moore titles “theory”, he presents a collection of one-sided opinion papers and reviews on listening difficulties, questioning the existence of a diagnostic APD *gold standard*. These papers define the gold standard approach as requiring a single test to diagnose a disorder. However, clinicians know that best clinical practice requires that developmental disorders and the majority of neurological diseases require more than a single test to reach an accurate diagnosis. Moreover, it is widely recognized that due to the complexity of disorders there is no single test or symptom that proves a diagnosis, which is why a battery of tests should be used in clinical practice (Jerger and Hayes 1976; Kasper et al. 2015). Thus, Moore’s point that since there is no single test to definitively diagnose APD, there is no gold standard, is moot. A gold standard for any given diagnosis is considered the best available and widely used method (Farlex Partner Medical Dictionary 2012). Currently,

the best available gold standard approach is included in the AAA (2010) guidelines that are also endorsed by several audiological societies and APD professional groups worldwide (e.g. International Bureau for Audiophonologie, 2007; Canadian Interorganizational Steering Group for Speech-Language Pathology and Audiology 2012; Auditory Processing Disorder: New Zealand Review 2014; Danish Medical Audiological Society 2014; Deutsche Gesellschaft für Phoniatrie und Pädaudiologie (DGPP) 2015; European APD Study group 2017). Going forward, we encourage research to refine the test battery, determine the most efficient test combinations (e.g., Burgueti & Carvallo 2008; Butler et al. 2011; Musiek et al. 2011; Allen and Allan 2014; Weihing et al. 2015; Barrozo et al. 2016), as well as develop innovative approaches, rather than ban scientific inquiry, as does *Ear and Hearing*'s new editorial policy.

Moore suggests that neurological underpinnings of APD are only or primarily seen in adults; however, he ignores the accumulating literature (some of which we cited above) that identify neuro-morphological or frank neurological lesions underlying APD in some children (e.g. Bamiou et al. 2007, Iliadou et al. 2008; Boscarriol et al 2011; Chermak and Musiek 2011; Koravand et al. 2017 and others cited above). Moore similarly omits several studies providing animal models for neurodevelopmental aspects of the CANS (Alexander et al. 2014a, b; Anomal et al. 2015). He also injects the suggestion that attention is the key substrate of APD; however, attention deficits are neither a uniform nor a core supramodal deficit in the majority of children diagnosed with APD (Stavrinos et al. 2018). A number of basic and clinical research findings argue against attention as the primary driver of APD (e.g. Dawes and Bishop 2008; Banai et al. 2011; Moore et al. 2011; Gyldenkaerne et al. 2014). Gyldenkaerne et al. (2014) reported that abnormal performance on central auditory tests often occurs despite sustained attention within normal limits and Banai et al. (2011) reported that maturation rates for different auditory tasks are not correlated, as would be expected if a nonsensory factor (e.g., attention) had a uniform influence on performance.

Moore criticizes the evidence underlying APD by lamenting that many publications on APD state opinions. Ironically, he cites several such publications throughout his editorial. For example,

one of the opinion papers he cited suggested that a child with confirmed communication and/or listening difficulties should only be *screened* for hearing and not diagnostically assessed as is best clinical practice (DeBonis, 2015). The arguments were based, however, on papers that involved groups of individuals suspected rather than diagnosed with APD. The problem with the “suspected APD” classification is underscored in a number of papers demonstrating that subjective report of listening problems is a poor indicator of actual performance-based auditory processing difficulties and fails to differentiate these from children with primary language disorders (e.g., Barry et al. 2015; Ferguson et al. 2011; Lam and Sanchez 2007). We would thus counter argue that if children with APD are not properly evaluated they might be erroneously labelled as having attention issues since the “diagnosis” would be based on questionnaires, thus depriving these children of appropriate auditory interventions.

APD Test Battery Norms

Moore criticizes studies that are based on original experimental data, arguing that they adhere to test batteries endorsed by professional organizations (e.g., AAA and ASHA) that he asserts, for the most part, do not have norms, and where they do are from sources “unavailable for verification.” There are, however, numerous publications describing norms either in the form of cut-offs for specific ages in children (i.e. Bellis 1996; Iliadou et al. 2009) or in the form of means and standard deviations (i.e. Shinn et al. 2009; Tomlin, Dillon, Kelly, 2014; Pedersen et al. 2017; Mattsson et al. 2018; Nickisch et al. 2018; Kiese-Himmel et al. 2018).

Prevalence of APD

Moore’s section on “clinical prevalence, presentation, and evaluation” begins by citing his retrospective record review in which 1113 children with normal audiograms had undergone a “central auditory processing evaluation” using the SCAN battery (Moore et al. 2018). Based on the SCAN, Moore concluded that only 14 of those children would be classified as qualifying for an APD diagnosis, but that all but one presented other diagnoses (i.e., speech-language, attention disorders, or

learning disorders) that provided an “alternative explanation of their listening difficulties.” Moore’s point is unclear since the SCAN is a screening rather than diagnostic battery, and given its language content is prone to a strong language confound.

Moore also cites his personal *unpublished, prospective study* (which is not available for evaluation of its methodological rigor) that only 2 of 100 children “extensively examined” using “the most specific and sensitive tests of central function” presented any type of “auditory-based problem.” However, he does not identify the test battery used, nor how the sensitivity and specificity of this battery were derived. His claim of 2 of 100 (2%) is thus no more than “anecdotal evidence”. Furthermore, the prevalence of 2% in his unpublished data set contradicts the 10% prevalence reported in a recent paper that he co-authored (Brewer et al. 2016) by a five-fold difference. Even if we assume that 2% of children more accurately reflects the prevalence of APD, it is unclear the point Moore is arguing. A 2 % prevalence for any condition is significant and even if APD were considered a rare disorder (with an occurrence of less than 1%), rare disorders are not to be ignored. Acoustic tumors, for example, occur far less than 2% of the time but we certainly do not ignore them. No matter the prevalence rate, our professional ethics and our moral compass demand that each child diagnosed with APD receive the best intervention available to optimize academic social and community access and quality of life.

Moore concludes this section of his editorial stating that children in his prospective, unpublished study presented poor “speech in noise hearing”, which “correlated with slight to moderate impairment of cognitive skills [using the NIH Cognitive Toolbox”, citing Weintraub et al. (2013)]. However, Weintraub et al. state that the NIH-Toolbox Cognition Battery was “not developed as a clinical measure to either screen for cognitive impairment or to substitute for a full, competent neurological evaluation.” (p. S61). Therefore, it is unclear how Moore determined slight to moderate impairment of cognitive skills in his sample using the NIH Cognition Toolbox. Clinical evaluations using standardized tools developed for research with undocumented efficiency for clinical diagnosis

is yet another reason why research in APD (as well as in any other disorder) must be encouraged not restricted.

Availability of APD Evaluation

Moore cites personal communication (not published and with unknown rigor) regarding a small informal survey to assert that there has been a decrease in the number of US children's hospitals offering APD evaluation to imply that this is because APD is not well defined nor measurable. The authors of this rebuttal who practice in the US would argue that *if* there is a diminution in services it is most likely due to: 1) poor third party reimbursement, 2) time required to complete a thorough evaluation, and 3) limitations in preparation in this area of practice in AuD programs. We reported these causative factors in a 2007 peer-reviewed publication (Chermak et al. 2007), and unfortunately, we are not aware of any evidence to suggest that these factors have changed substantially. However, the recent inclusion of central auditory processing disorders in the ICD-10 may provide the impetus for improved reimbursement by U.S. insurance providers, as is the case in other countries around the world when those claims are coded consistent with the ICD-10.

Listening Difficulties v. APD

To our astonishment, Moore suggests that “listening difficulties” may be a better umbrella term to capture the range of deficits that ultimately impact listening—presumably including APD, attention, memory, and language—and could serve as a “diagnostic billing category.” In fact, listening difficulty is a symptom; APD is a disorder (ICD-10 & ICD-11 Beta version). Listening difficulties may be seen in autism, hearing impairment, dyslexia, attention deficit hyperactivity disorder, and numerous other disorders, and as such listening difficulties is a nonspecific term that contributes little to differential diagnosis. As argued above, this is why questionnaires or self-report can lead to incor-

rect (over or under) diagnosis of APD. The overlap of symptoms across different disorders is particularly evident in (1) neurodevelopmental disorders and (2) those included in the DSM-5. We are stunned that Moore would suggest the term ‘listening difficulties’ to replace APD when many children referred for APD evaluations because of “listening difficulties” actually perform quite well on central auditory processing measures (Sharma et al. 2009; Rosen et al. 2010).

A thorough evaluation relying on central auditory processing tests is mandatory to diagnose APD. APD is a complex disorder and it is essential that the clinician (audiologist) responsible for diagnosing the disorder consult with other members of a multidisciplinary team to ensure accurate differential diagnosis and identify comorbid disorders to maximize efficient and effective intervention. Such treatment and management must be individualized and targeted to the deficit-specific processes identified by the diagnostic test battery (e.g., Cameron and Dillon 2011). Accumulating evidence has documented the success of auditory interventions following an APD test battery of documented sensitivity and specificity (see Filippini et al. 2019 and Weihing et al., 2015 for reviews).

What We Can Learn from the DSM-5

One of the reasons the DSM was updated (DSM-5) was the realization that “the once plausible goal of identifying homogeneous populations for treatment and research resulted in narrow diagnostic categories that did not capture clinical reality, symptom heterogeneity within disorders, and significant sharing of symptoms across multiple disorders.” (DSM-5 introduction page 12, lines 31-34). The way forward is well stated in the DSM-5’s cautions that : “...a too-rigid categorical system does not capture clinical experience or important scientific observations” and that “... the boundaries between many disorder “categories” are more fluid over the life course.” The DSM-5 argues that the fluidity of boundaries across disorders “should permit a more accurate description of patient presentations and increase the validity of a diagnosis” (DSM-5 page 5, 25-27, 29-30, 34-35). The authors

of this rebuttal fully support the DSM-5 approach, an approach which is the antithesis of *Ear and Hearing*'s editorial decision to not publish research on APD due to its diverse clinical presentations, overlapping symptoms, and comorbidities.

Call to Action

Moore concludes his editorial by declaring again (as he did in his introduction) that the *Ear and Hearing* Editorial Board has decided that “articles that either implicitly or explicitly assume APD is a single diagnostic characteristic of the auditory system likely will not be considered for publication.” Since APD is included in the ICD-10 and the ICD-11 Beta version as a specific type of hearing impairment/ear disease for both acquired and congenital APD-- *Ear and Hearing*'s decision is indefensible. *Ear and Hearing*'s editorial decision attempts to restrict and diminish scientific inquiry and research and slow advancement of our understanding, diagnosis, and treatment of APD, by excluding publication of research based on a narrow and predetermined concept of what APD is or is not rather than on the merits of the research methodology (i.e. the collection, measurement, analysis, and clarity of presentation of data). This contradicts all tenets of the scientific process.

Carl Sagan once said that valid criticism does you a favor. We would hope that constructive criticism rather than a biased dismissal and banning of rigorous research would be fundamental policy of a journal that purports to serve the interests of the broad audiological community. Some of us have played a role in the development of *Ear and Hearing* from its inception. We have published and reviewed numerous articles for this journal. Over the years, we have advocated for *Ear and Hearing* in many ways. Given this history, we are indeed disappointed. We reject this editorial policy decision. We see no evidence that Moore's editorial was properly vetted, with the thoroughness and fairness that has always been a hallmark of *Ear and Hearing*'s editorial values. It appears that this policy decision has been rendered without any discussion or input from those with different perspectives. If this editorial policy stands, it will be a serious affront to open, rigorous scientific debate and broad-

based, cross-disciplinary representation that *Ear and Hearing* has championed since its first publication in July of 1975. Moore notes that he has reviewed hundreds of submissions on APD. We wonder how many scientifically worthy publications were rejected due to his bias.

Based on the foregoing, and unless and until this editorial policy is reversed, the coauthors of this rebuttal--- composed of researchers and clinicians with considerable experience evaluating and treating individuals with APD--- will no longer submit manuscripts to *Ear and Hearing* and will encourage others to refrain as well due to *Ear and Hearing*'s restriction on scientific inquiry.

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