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Review Article

Contentious issues related to Auditory Processing Disorder

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1. Introduction

A number of children undergo audiometric evaluation due to suspected hearing difficulties. Yet for some, their hearing sensitivity is found to be normal (Iliadou et al., 2017; Sharma, Purdy, & Kelly, 2009). Often, these children are described by parents and teachers as having difficulties to follow several instructions or attend to a conversation, and getting distracted when background noise is present (Iliadou et al., 2017; Witton, 2010). There are also reports of these children either misunderstanding or taking longer to understand simple verbal directions, and demonstrating selective hearing (Johnson, Benson, & Seaton, 1997). These weaknesses often cause educational, vocational and social challenges (American Speech-Language-Hearing Association (ASHA), 2005; British Society of Audiology (BSA, 2007). Children exhibiting these problems could be diagnosed with Auditory Processing Disorder (APD) (American Academy of Audiology (AAA), 2010; ASHA, 2005; BSA, 2007; Iliadou et al., 2017).

2. What is auditory processing (AP)?

The hearing process is not simply the transduction of acoustic signals into neural impulses within the ear (Bamiou, Musiek, & Luxon, 2001). These impulses are then transmitted via the auditory nerves to the brain. AP is the proficiency of the central nervous system to process

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information originating from the auditory channels. It incorporates a system of electrophysiological auditory potentials and involves both sound detection and its transmission to the brain through the auditory pathways (Yalçinkaya, Muluk & Şahin, 2009). The conscious perception of speech and non-speech auditory signals takes place in the auditory cortex: the site where bottom-up and top-down processing come together (Moore & Hunter, 2013). Several studies have suggested likenesses and overlap between auditory and speech processing (e.g. Benasich et al., 2006; Joanisse & Gati, 2003; Zaehle, Meyer, & Jäncke, 2004).

There are two leading hypotheses underlying deficits in AP. One hypothesis, the pathway model of AP, proposes that these deficits originate from impaired bottom-up sensory processing, made up of the ear and the central auditory nervous system (CANS). This hypothesis describes it as being sound driven (Wilson, Heine, & Harvey, 2004). The bottom-up approach emphasis the manipulation of sounds at different levels of the CANS. It implies that the sound properties regulate higher-level representations (Chermak & Musiek, 1997). Bottom-up processing deficits within the auditory nervous system could affect sound localisation, auditory discrimination, temporal processing, and auditory performance in the presence of competing and degraded sounds (Wilson, 2018).

The other hypothesis (the network model of AP (Wilson et al., 2004)) contrasts with the pathway model, implying that auditory processing is knowledge- or concept-driven, so that the sound processing is regulated by higher-level processing and sounds are then interpreted accordingly (Chermak & Musiek, 1997). AP weaknesses thus result from top-down effects of cortical

cognitive processing, which would in turn influence language processing, attention and memory, and indirectly affect auditory perception (Moore & Hunter, 2013). The network model highlights the combination of sound, meaning and intention and goes beyond the auditory pathway (Friel-Patti, 1999).

Although bottom-up and top-down theories differ in their approach to auditory processing, they are not exclusively contradictory (Friel-Patti, 1999; Moore & Hunter, 2013). The combination of both functions supports the processing of auditory information (Bellis, 2003). While bottom-up processing is fundamental to auditory perception, providing information related to incoming sounds, top-down influences are essential to regulate the incoming signal through the integration of sounds with an individual's experiences and expectations (Moore & Hunter, 2013). This in turn aids bottom-up processes to be informed of both new auditory signals and data that are mismatched with a current hypothesis about that sound (Wilson, Heine & Harvey, 2004). This complex interrelation between bottom-up and top-down pathways could pose difficulties to unravel each process in a clinical assessment (Wilson et al., 2004; Moore & Hunter, 2013).

Defining auditory processing disorder (APD)

APD has progressively gained recognition in audiology over the last couple of decades (Ludwig et al., 2014). It has been described as a combination of unrefined listening skills that cause difficulties with speech perception. It is evident especially in noisy environments, which challenge the individual further (Rosen, Cohen & Vanniasegaram, 2010). These difficulties are evident despite normal hearing levels detected on standard audiometry (de Wit et al., 2016).

In the ASHA (2005) technical report, APD has been described as a disorder in the perceptual processing of both speech and non-speech sounds in the CANS, resulting in weak performance in one or more of the following skills: discrimination of auditory information, recognition of auditory patterns, temporal processing skills, auditory performance in the presence of competing sound stimuli, and auditory performance when acoustic information is degraded. The degree to which this perception is affected depends on the specific processes that are weak as well as the complexity of the auditory information to be processed (Price, Thierry, &

Griffiths, 2005). The ASHA report further acknowledged that although the definition of APD describes the most noticeable difficulties in sensory *auditory* processing, the fact that sensory processing in the central nervous system is supported by language and cognitive skills should not be excluded. This was shown in an number of electrophysiological studies (e.g Bajo, Nodal, Moore, & King, 2010; Clark, Rosen, Tallal, & Fitch, 2000; de Boer & Thornton, 2008; Irving, Moore, Liberman, & Sumner, 2011; Tallal, Merzenich, Miller & Jenkins, 1998), and tends to cause ambiguity in the understanding as to *what* should be incorporated into APD and what clinical assessments should be administered to obtain a differential diagnosis.

The BSA's position statement of 2011(b) and 2018 suggests that APD includes the reduced perceptual ability of both non-speech and speech sounds, which in turn causes poor listening abilities. It recommends that APD is diagnosed through non-speech tests along with the speech-based tests. Thus, if an auditory deficit is present solely in speech processing or phonological categorisation, it is not considered as APD. However, with research (Moore et al., 2010, Watson & Kidd, 2009) disclosing no significant or consistent link between results on simple stimuli non-speech psychoacoustic tasks and the listening difficulties described by subjects, this approach on APD might not present itself as a good association between the purpose for referral and the diagnostic assessment (Moore, Rosen, Bamiou, Campbell, & Sirimanna, 2013).

The BSA (2018) definition states that APD originates from an impairment of neural function within the afferent and efferent pathways of the CANS, together with the related top-down modulation (vision and the cognitive functions of speech and language, attention, executive function, fluid reasoning, memory and emotion). Therefore, APD would often co-occur with (and could contribute to) the primary disorders of those systems. The BSA (2018) reports that "APD may thus include both auditory and cognitive elements" (p. 6). The European consensus (Iliadou et al., 2017) on APD is in agreement with the BSA definition to an extent. The authors however regard APD as an entity on its own. In cases where APD is comorbid with cognitive deficits, they stress the importance of differentiating APD from other neurodevelopmental disorders, through scientific evidence-based agreement regarding the direction of causality or the shared underlying pathology. With the complex interlinking of language, cognition and AP,

and the influence which each could have on another, a multidisciplinary approach could exclude the influence of a higher order disorder on the AP result.

These variations between APD definitions make it unclear as to where to draw the line between an auditory and language disorder. The evidence of this complex processing has led researchers to query whether it is necessary to adopt an APD diagnosis which is completely specific to the auditory modality (DeBonis & Moncrieff, 2008). Cacace and McFarland (2005, 2013) propose that a definition based on 'modality-specificity' could avoid the ambiguity of what is (or not) APD. The authors define it as a modality-specific perceptual dysfunction which is not brought about by peripheral hearing loss, and is distinct from similar difficulties arising from impairments in cognitive, language, and/or attention skills. The argument put forward by these authors has been questioned, with suggestions to define APD as primarily modality specific rather than exclusively specific to the auditory perceptual modality (Musiek, Bellis, & Chermak, 2005).

4. Prevalence of APD

Prevalence reports on APD are diverse. Studies show prevalence estimates in the paediatric population ranging between 2 and 10% (Bamiou, Musiek & Luxon, 2001). More recent findings suggest lower percentages. A study on 243 children in the United States found a paediatric prevalence of 0.2% (Nagao et al., 2016), while in the United Kingdom a prevalence of 0.5% was suggested (Hind et al., 2011). When APD is present together with other learning disabilities, its prevalence has been reported to increase to between 30 and 50% (King, Warrier, Hayes & Kraus, 2002; Ramus, 2003). Despite this variation, APD is currently included in the International Classification of Disorders version 11 (ICD-11) (World Health Organisation (WHO), 2018).

5. Comorbidity of APD

The characteristics frequently associated with APD can overlap with other developmental disorders. One presenting factor is poor attention and distractibility (ASHA, 2005; BSA, 2011b; Jerger & Musiek, 2000; Riccio, Cohen, Garrison, & Smith, 2005; Witton, 2010). According to the diagnostic manuals by the WHO (ICD-10) (1993) and the American Psychiatric Association (APA) (DSM-V)

(2013), this symptom is also dominant in children with attention deficit hyperactivity disorder (ADHD).

5.1. ADHD

ADHD is a developmental disorder generally evident by the time a child starts primary school. It is characterised by persistent inattention. These individuals may also exhibit hyperactivity/impulsivity, which could interfere with their development or functioning (APA, 2013). Considerable work has been put into investigating the comorbidity of ADHD with other developmental disorders (Pliszka, 2009), though this differentiation can be challenging (Sulkes, 2013). The presenting symptoms of ADHD and APD are associated with poor auditory attention, listening skills, and academic performance (Chermak, Tucker & Seikel, 2002). Rosen, Cohen and Vaniasegaram (2010) found that 60% of the children suspected of having APD also reported difficulties with attention and concentration. However, research has also suggested that while deficits in AP and sustained attention can co-occur in some children diagnosed with APD, the two conditions are distinct (Gyldenkærne, Dillon, Sharma & Purdy, 2014). These overlapping symptoms result in ambiguities with differential diagnosis (Brown, 2009). So, exploring AP in children with ADHD has become of interest.

Research on Maltese children compared the listening skills in children with ADHD to typically those in developing children via a parental questionnaire (Tabone et al., 2016). The clinical group was found to perform significantly worse than the typically developing children in auditory attention and memory, conversation skills, sensory stimulation, listening in noise, and situations involving social interaction. This study also compared the performance of the two groups on behavioural AP subtests using both speech and non-speech stimuli. The ADHD group performed significantly worse than the controls on tests of dichotic listening (using speech stimuli). Analogous outcomes were reported in other studies (Dramsdahl, Westerhausen, Haavik, Hugdahl & Plessen, 2011; Manassis, Tannock & Barbosa, 2000), where authors attribute this to a cognitive control deficit in conflict situations.

Reports on the effects of noise in individuals with attention difficulties have been conflicting. Some studies report better or at par performance by individuals with attention difficulties on speech-in-noise tests (e.g. Söderlund, Sikström, & Smart, 2007; Söderlund,

Sikström, Loftesnes & Sonuga-Barke, 2010; Söderlund & Jobs, 2016; Tabone et al., 2016). These studies suggested that background noise may actually be beneficial for cognitive performance in children with attention deficits. Other studies have reported significantly poorer performance in children with ADHD on tasks of speechin-noise (Abdo, Murphy & Schochat, 2010).

Research on ADHD theory proposes that weaknesses in the processing of temporal information could contribute to poorer cognitive and behavioural results. However, methods of testing temporal skills differ and could make it difficult to compare findings across studies (Toplak, Dockstader & Tannock, 2006). There seems to be agreement that individuals with ADHD do not show significant deficits in frequency temporal processing (Abdo et al., 2010; Radonovich & Mostofsky, 2004; Tabone et al., 2016; Toplak, Rucklidge, Hetherington, John & Tannock, 2003). However, conflicting outcomes are reported on performance in duration temporal processing tasks (e.g. Radonovich & Mostofsky, 2004; Van Meel, Oosterlaan, Heslenfeld & Sergeant, 2005).

5.2. Language and literacy

Difficulties with language and/or communication are also frequently reported in children diagnosed with APD (ASHA, 2005; BSA, 2011; Ferguson, Hall, Riley, & Moore, 2011; Sharma, Purdy, & Kelly, 2009). The relationship between APD and language/literacy deficits is possibly the most debated and discussed in the literature (Dawes & Bishop, 2009). One theory suggests that a language deficit emerges due to impaired auditory perception (Feldman & Messick, 2009). This was initially proposed by Tallal and Piercy (1973). Their study, investigating the temporal processing skills in children, showed that some children with phonological difficulties found it problematic to detect quick temporal changes in sounds. This could lead to weak literacy skills, particularly in phonemic awareness. There were additional early studies supporting Tallal and Piercy's (1973) claim (e.g. Lubert, 1981; Tallal, 2000; Tallal, Miller, & Fitch, 1993; Tallal & Stark, 1981). Frumkin and Rapin (1980) showed that children with a language disorder and additional phonological difficulties had a temporal processing deficit, while those who did not have phonological difficulties did not. Some studies have suggested that deficits in auditory perception of amplitude and pitch variation could cause difficulties perceiving prosody, and consequently in interpreting the meaning of spoken phrases (Bellis & Ferre 1999; Griffiths, Johnsrude, Dean, & Green, 1999).

Other studies have further investigated the earlier claims of temporal processing as an underlying cause of deficits in language and/or literacy, with controversial outcomes. Some studies concluded consistent (or partially consistent) results with Tallal's temporal processing hypothesis (e.g. Cantiani, Lorusso, Valnegri, & Molteni, 2010; Cohen-Mimran & Sapir, 2007; Groth, Lachmann, Riecker, Muthmann, & Steinbrink, 2011; Heiervang, Stevenson, & Hugdahl, 2002; Schulte-Körne, Deimel, Bartling, and Remschmidt, 1999). However, there have also been studies that failed to observe similar findings (e.g. Breier, Fletcher, Foorman, Klaas, & Gray, 2003; Bretherton & Holmes, 2003; Mody, Studdert-Kennedy, & Brady, 1997; Ramus, 2004; Watson et al., 2003). Sharma et al. (2006) combined both behavioural and electrophysiological measures to investigate the AP skills in children with literacy difficulties. Their findings indicated that these children can exhibit poor frequency pattern discrimination as well as speech-syllable discrimination deficits.

Therefore associations between the auditory and language/literacy impairments may stem from common developmental substructures, rather than causality (Witton, 2010). Conflicting results on the association between the disorders have been reported. There has been research suggesting that weaknesses in AP skills are not necessarily related to speech, language and literacy deficits (Hazan, Messaoud-Galusi, Rosen, Nouwens, & Shakespeare, 2009; Ramus, White, & Frith, 2006; Rosen, 2009; Watson & Kidd, 2009). For example, Watson et al. (2003) found that AP of speech sounds in the presence of difficult listening conditions was a poor predictor of academic achievement. Hazan et al. (2009) also failed to find a link between phonological processing and speech perception in children with dyslexia. Rosen, Adlard, and van der Lely (2009) analysed performance of children with grammatical language difficulties when presenting tones both in quiet and in background masking noise. They reported a poor correlation between this nonspeech AP task and measures of vocabulary, grammar, and phonology in these children, and suggested that the deficits in AP skills sometimes present in children with language disorders seem unlikely to be the cause.

In contrast, there have also been studies showing comorbidity between auditory processing disorders, and disorders of language and literacy. Sharma, Purdy, and Kelly (2009) investigated the auditory, language, reading,

attention, and memory abilities in children suspected of having APD. Their results indicated that while 72% came out with a profile of APD, only 4% resulted to have solely difficulties with AP. Nearly half of the children showed weaknesses in all areas of auditory, language, and literacy skills, and a larger number resulted to have APD comorbid with language or literacy deficits rather than a single deficit. Co-morbidity between these disorders has also been investigated by means of electrophysiological measures. One study (Weber-Fox, Leonard, Hampton Wray & Tomblin, 2010) used both non-speech and speech-based stimuli to examine the neural activity for rapid auditory processing in adolescents with a language disorder. The findings resulted in atypical AP of both the tonal and speech-based stimuli, suggesting a possible overlap between APD and language disorder.

An explanation for these inconsistent results could be the variation within the population of children with language - related disorders (Dawes & Bishop, 2009), where some sub-groups may have additional or underlying deficits with AP and other sub-groups may not. The conclusions of these studies have led researchers to investigate the likelihood of a language disorder arising because of a deficit in higher order processing rather than poor auditory perception. Perhaps one way of exploring the link between AP and language disorder is to look into the possible co-morbidity within groups not diagnosed with a language disorder, but with a different pathology. Halliday, Tuomainen and Rosen (2017) attempted to do this by assessing the AP and language skills in individuals with no language disorder, but with a mild to moderate sensorineural loss (also known to exhibit problems with AP skills). Their study suggests several routes that associate AP weaknesses with language deficits, rather than a specific AP deficit causing language difficulties. It concludes that AP deficits might (but not always) be necessary and enough to result in a language disorder.

6. Assessment of auditory processing skills

There is a lack of agreement in the acceptance of a 'gold standard' for diagnosing APD. This results in substantial variability among centres who assess for APD (BSA, 2011a) and problems meeting the conditions necessary to develop a robust tool (Keith, 2009): having good construct validity (Johnson, Bellis, & Billiet, 2007) and test-retest reliability (Cacace & McFarland, 2005), a high sensitivity

and specificity (Wilson & Arnott, 2013), standardisation (Dawes & Bishop, 2009) and cut-off scores (Keith, 2009) for each of the tests in specific population groups.

6.1. Differential diagnosis of APD

Clinicians working with individuals queried for APD in their daily routine, have been requesting guidelines to aid with the best management. This has stimulated several research studies to search for strong evidence of the disorder and symptoms it exhibits, so that the most suited diagnostic criteria and intervention strategies are recommended (BSA, 2011a). Its differential diagnosis is particularly important because of the findings that APD may co-exist with other disorders (Dawes & Bishop, 2008; Ferguson & Moore, 2014; Ferguson et al., 2011; Miller & Wagstaff, 2011; Sharma et al., 2009; Witton, 2010), warranting the importance of teasing out the difficulties specific to the auditory modality.

The differential diagnosis of APD is still much debated, with disagreements on the underlying theoretical model and the availability of a 'gold standard' for its diagnosis. Iliadou, Chermak, Bamiou and Musiek (2019a) suggest that the auditory processing test battery approach, discussed later in this paper, is the most suited gold standard approach in the diagnosis of APD, since it is the best evidence-based diagnostic methodology available (Iliadou et al., 2017). Their proposed APD gold standard was equated with that of the pure tone audiogram gold standard of diagnosing hearing loss (Iliadou et al., 2019a). This approach has been questioned, with statements that the traditional auditory assessments may lack evidence, posing difficulties to differentially diagnose APD (Neijenhuis et al., 2019). However, recent proposals emphasise that APD can be accurately diagnosed through the use of clinical expertise as well as the available research and patient values (Iliadou et al., 2019b). Diagnosing a disorder is a process that depends on symptoms. Test findings should involve the recognition of a specific pattern by the diagnosing clinician. It is an iterative process, by which clinical hypothesis is formulated on the basis of the patient's presentation and then confirmed or discarded on the basis of a range of findings and additional information (Kohn, 2014).

6.2. The AP test battery approach

In light of this controversy, a multidisciplinary assessment is recommended by audiological associations, involving: (1) in-depth audiometric testing in order to detect peripheral hearing loss and distinguish it from other disorders with similar presenting symptoms, such as Auditory Neuropathy Spectrum Disorder (ANSD); (2) an assessment battery of auditory processing (BSA, 2007). The AAA (2010) suggests primarily behavioural AP assessments. The Canadian Interorganizational Steering Group for Speech-Language Pathology and Audiology (CISG, 2012) further emphasises the use of a behavioural questionnaire prior to commencement of the assessment battery, where information related to the individual's listening skills is obtained from caregivers, speech-language pathologists and educators. This could help expose the functional impact of the APD, and determine whether further assessment of AP is necessary. Questionnaires can guide the audiologist to the choice of tests to be administered, and subsequently their interpretation. However, the questionnaires used may not always be predictors of the APD assessment outcomes (Tabone, 2018) and therefore it is suggested that they are not used alone to determine the presence of APD (Wilson et al., 2010).

Audiological associations (e.g. BSA, 2018; AAA, 2010; ASHA, 2005) recommend that an AP assessment battery includes both speech and non-speech auditory stimuli. The tests should also target dysfunction of the different neuroanatomical regions along the CANS (Johnson et al., 2007), such as dichotic listening tests, temporal processing and patterning tests, artificially degraded speech, binaural interaction, as well as the use of electrophysiological measures. The disadvantage of this approach might be that an increased amount of tests could lead to a greater chance of a child performing badly on one, and misinterpreted as a deficit of the entire assessment battery (Dawes & Bishop, 2009). Multidisciplinary assessment of speech and language (Bamiou, Campbell & Sirimanna, 2006), auditory memory and attention (BSA, 2011a) has been recommended in light of research studies indicating possible overlap between these impairments (Rosen, 2009). This could in turn cause a misdiagnosis and incorrect management of these children (BSA, 2011a). Clinical observation across different listening environments has also been emphasised (Bamiou et al., 2006). In a survey completed by 195 audiologists in the United States, it emerged that before administering an AP assessment battery, 33% of the audiologists carry out a classroom observation, where children might encounter greater difficulties (Emanuel, Ficca & Korczak, 2011).

Dillon, Cameron, Glyde, Wilson and Tomlin (2012) consider APD through a different perspective. These authors emphasise the importance of investigating the difficulties understanding *speech* in challenging listening conditions, and suggest an AP assessment which adopts a hierarchical approach focused on listening difficulties linked to speech situations.

ASHA (2005) recommends either a poor performance (of two standard deviations below the mean) on at least two sub-tests in order give a diagnosis of APD, or a very weak performance of three standard deviations below the mean on one sub-test. Although the BSA (2011a, 2018) does not specify any diagnostic criteria, it highlights the poor perception of both speech and non-speech sounds. Therefore an individual must perform poorly on two sub-tests, one speech-based and one non-speech-based test, in order to be diagnosed with APD (Wilson & Arnott, 2013).

Several aspects should be considered when compiling and administering an APD assessment battery. One is the age of the children to be assessed. While there have been studies investigating AP skills in very young children (e.g. Sidiras, Iliadou, Chermak, & Nimatoudis, 2016; White-Schwoch et al., 2015; Stephen et al., 2012), many behavioural assessments suggest that testing starts when children reach 7 years of age (e.g. Mattsson et al., 2018; McDermott et al., 2016). This is due to the increased variability of brain function in younger children, resulting in difficulties to interpret results (Whitelaw & Yuskow, 2006). Maturational effects in the CANS have even been recorded until children reach approximately 12 years of age (Moore et al., 2010). Young children may find it difficult to understand and follow task directions, which may potentially lead to unreliable results. Similarly, when assessing children with diagnoses of other developmental disorders, their developmental age, language and cognition skills should be taken into consideration since they could be functioning at a similar level to younger children (Fong, 2016). The time factor is also important for clinical practice. The administration of an assessment battery should not take too long, especially when the sub-tests are demanding. Children usually have a relatively short attention span and tend to fatique quicker. Thus an assessment battery that is complete in not more than 60 minutes is desirable (AAA, 2010). Repeated breaks and consistent reinforcement

may also help keep children motivated to complete each task.

7. Conclusion

Studies have suggested that when a child is diagnosed with a developmental disorder, there is a significantly increased chance that the symptoms of other developmental disorders are also present (Witton, 2010). This may be attributable to the extent with which the brain regions are interconnected, so that cognitive sections like language and memory do not develop and function independently, but rather interact through complex processes (Karmiloff-Smith, 1998). It seems to be the main present focus of researchers on whether APD can manifest itself independently of other disorders, i.e. as a 'pure' disorder, or whether it is a characteristic present as part of a developmental disorder. Should the causal mechanism of APD be of research interest, then it would warrant the investigation of individuals presenting specifically with a 'pure' disorder of APD and reducing or removing any additional deficits that may confound the results (Dawes & Bishop, 2009). Nonetheless, this population may not be highly prevalent (Witton, 2010). However, this direction of research may not be entirely significant for clinical purposes, where many of the children may also have the deficits in hearing, language, literacy, attention, and social skills (Dawes & Bishop, 2009). Research studies concerned with the clinical aspect of APD tends to be more focused on the link between APD and the other disorders. The debate is ongoing regarding whether the auditory deficit is the primary cause or a secondary consequence of the other difficulties (Dawes & Bishop, 2009), or whether these developmental disorders simply co-occur (BSA, 2011a,b). In an attempt to settle this dispute, the concept of merging the different approaches, definitions and positions to APD into an AP spectrum disorder has been proposed (Wilson, 2018), where a variety of related conditions with similar appearance or sharing the same underlying mechanism would be included. With the complex link and interrelationship between AP, language and cognition, clinical management of an AP spectrum disorder could target the clinical presenting deficit of every individual child.

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Conflicts of interest

The authors report no conflicts of interest.

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