Listening difficulties in children

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GENERAL INTRODUCTION
INTRODUCTION

Learning new knowledge and skills are important human abilities. Much of what we learn as human beings takes place at primary school. To learn, a child must develop academic skills, such as communication skills, for example listening and adequately processing of and responding to non-verbal and verbal information, and cognitive skills, such as concentration, attention and memory. From the age of six, the capacity to process information markedly increases and the control of attention and memory improves (Verhulst, 2017).

Normal hearing is essential for the acquisition of oral language and effective verbal communication (Bhatnagar & Korabic, 2006). Most children will learn without problems, but some children experience learning difficulties. One of the learning difficulties that may occur in children, is having problems with listening and processing auditory information, despite normal peripheral hearing ability. Children which such problems, have difficulties with understanding speech in complex listening situations, such as in a busy classroom with a lot of background noise. These children are described by their parents and teachers as children who are uncertain about what they hear, have trouble listening in the presence of background noise and have difficulty in following oral instructions. They have trouble understanding rapid or disturbed speech and are unable to follow conversations and are inattentive (Jerger & Musiek, 2000; Hind et al., 2011; Moore, Rosen, Bamiou, Campbell & Sirimanna, 2013).

Problems in auditory processing are complex and not yet fully understood. It is unclear whether auditory processing difficulties should be regarded as a pure auditory disorder with an underlying etiological unity that is distinct from other learning disabilities or that the listening difficulties are caused by deficits in the field of intelligence, language, reading and spelling, attention and/or concentration (Moore, 2006; Dawes & Bishop, 2009; Miller & Wagstaff, 2011).

The term “listening difficulties (LiD)” is used to summarize the problems with hearing or listening, in spite of normal audiometry. These difficulties are typically reported by the caregiver or professional of the child and refer to the symptoms perceived at the child. The cause of the symptoms of these listening difficulties is not yet known. The term “listening difficulties” does not imply any underlying mechanism and cannot be used as a diagnostic label (Moore, 2018). In many audiology services around the world, the term "auditory processing disorder (APD)" is the clinical label or diagnosis used to classify the listening difficulties of children (Moore et al., 2013). The diagnosis APD refers to problems with the auditory processing of speech in everyday life in individuals who have normal hearing pure tone sensitivity. A description that is often given for APD is “when something goes wrong with what we do with what we hear” (Katz, Stecker & Henderson, 1992). Recently, APD is included as a disorder in the 10th version of the International Classification of Diseases as H93.25 and in the forthcoming beta 11th version (Iliadou et al., 2017).

However, not in every country the diagnosis APD is used to classify the problems of children with listening difficulties. This is due to the uncertainties surrounding the underlying cause of the listening problems in children and to the potential overlap between the symptomatology of APD and the symptoms of other neurodevelopmental disorders, such as developmental language disorder (DLD) or specific language impairment (SLI), dyslexia, attention deficit (hyperactivity) disorder (AD(H)D), and autism spectrum disorder (ASD) (Bamiou, Musiek & Luxon, 2001; Rosen, 2005; Hind, 2006; Moore, 2006; Cacace & McFarland, 2009).
Auditory Processing

Auditory processing refers to the processing of auditory stimuli by the outer ear, via the middle ear, to transduction in hair cells in the inner ear, up to and including the central pathways (brainstem, thalamus and cortex). When hearing a sound or spoken message by the peripheral auditory system, the meaning of the auditory stimulus, usually with a linguistic message included, must be processed by the central auditory system (CAS), so called central auditory processing. Central auditory processing refers to how the auditory sensory input and acoustic information from the environment is perceived and processed by the auditory pathway after it leaves the peripheral auditory structures (outer, middle, and inner ear) and what happens to this information as it is transmitted along the central auditory system (CAS) (Bailey, 2012; McNamara & Hurley, 2015; Perigoe & Paterson, 2015). Central auditory processing includes the auditory mechanisms that underlie the following skills: sound localization and lateralization; auditory discrimination; auditory pattern recognition; temporal aspects of audition, including temporary integration, temporally discrimination (e.g., temporal gap detection), temporal order and temporal masking; auditory performance in competitive acoustic signals (including dichotic listening); and auditory performance with degraded acoustic signals (American Speech-Language-Hearing Association (ASHA), 1996; Musiek & Chermak, 2007; Bellis, 2011).

Figure 1 illustrates the schematic representation of the central auditory pathway. The central auditory pathway consists of an extensive network of interconnected nuclear complexes in the brainstem and thalamus, and numerous areas in the cerebral cortex (Hackett, 2009). The auditory signal is transmitted via the cochlea to the auditory nerve and then transported by the auditory pathway through the brainstem, via the superior olivary complex that receives bilateral projections from the cochlear nuclei and where the auditory signal is converged, via the inferior colliculus (midbrain) and medial geniculate nucleus (thalamus) to the auditory cortex. This is the so called afferent or ascending pathway. The ascending pathway receives significant input from the efferent or descending pathway (Winer, 2005). Equally massive and equally specific descending projections take place from the auditory cortex to the medial
geniculate nucleus, inferior colliculus, superior olivary complex, cochlear nucleus, pons and basal ganglia. These descending pathways can affect many aspects of subcortical performance, including filtering, sharpness of tuning, and response plasticity (Winer, 2005).

**Bottom-up and Top-down Processing**

In order to be able to listen and understand what is being said, a child must detect the speech sound, focus on the speech sound, determine where the speech sound comes from, and separate it from background noise. Hearing and bottom-up auditory processing of the sound alone is not sufficient for interpreting and understanding the auditory stimulus. Various skills, such as guided attention, memory and language skills, that is, the so called top-down cognitive processes, play a role in correctly interpreting and understanding the message (Bellis, 2011; Moore, 2012;). In the ultimate processing of auditory input, both bottom-up factors (sensory encoding) and top-down factors (cognition, language and other higher-order functions) work together (Bellis, 2011). Even the simplest auditory signals are influenced by higher cognitive factors, such as memory, attention and learning (Mülder, Rogiers & Hoen, 2007). Thus, in addition to a well-functioning peripheral hearing and well-processing of the auditory signal, listening requires focus to the speech stimuli and the involvement of memory, intelligence and language skills (British Society of Audiology (BSA), 2007). Therefore, the processing of auditory information comprises both bottom-up and top-down processing. According to Moore (2006) it is difficult to clearly understand which role bottom-up and top-down processes play exactly when listening, since both processes contribute to almost all aspects of processing auditory information.

**Relationship between APD and other neurodevelopmental disorders**

One of the current pressing issues is whether APD is a unique clinical entity which can be regarded as a unimodal auditory-specific disorder or whether the listening problems are related to or caused by another impairment, for example, language- or attention difficulties (e.g., Bellis & Ferre, 1999; Cacace & McFarland, 2009; Dawes & Bishop, 2009; Ferguson, Hall, Riley & Moore, 2011; Kamhi, 2011; Miller & Wagstaff, 2011; Richard, 2011). It has been suggested that a child may receive the diagnosis APD, SLI, dyslexia or ADHD depending on the referral route of a child with reported listening difficulties. (Dawes & Bishop, 2009; Ferguson et al., 2011; Moore et al., 2013). This is obviously related to the lack of clarity on the causes of listening difficulties in children (Rosen, 2005; Cacace & McFarland, 2009), and its exact relationship with other neurodevelopmental disorders (Dawes & Bishop, 2009; Miller, 2011). Listening difficulties are associated with other neurodevelopmental disorders, but the exact relationships are unclear. The possible relationships are shown in Figure 2 (based on Miller & Wagstaff, 2011):

1. The listening difficulties in children are caused by a pure APD and the various neurodevelopmental disorders are distinct constructs and identifiable disorders that can be distinguished theoretically and clinically.
2. APD, SLI, dyslexia and ADHD are different labels for the same construct.
3. Listening difficulties are caused by language, reading, attention and/or concentration impairments and are a subset or symptom of one of the other neurodevelopmental disorders.
4. All other neurodevelopmental disorders are caused by APD and the symptoms of other neurodevelopmental disorders are a subset of APD.
5. A larger processing deficit or multimodal or general neurodevelopmental disorder exists in which the behavioral difficulties of children (e.g. auditory, language and attention) serve as indicators that can be expressed along a continuum of severity.
### Figure 2. Five possible relationships between the construct auditory processing disorders (APD) or listening difficulties (LiD) and other neurodevelopmental disorders (specific language impairment (SLI), dyslexia, and attention deficit hyperactivity disorder (ADHD)). (Based on the figure of Miller & Wagstaff, 2011).

<table>
<thead>
<tr>
<th>1. APD</th>
<th>SLI</th>
<th>Dyslexia</th>
<th>ADHD</th>
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<tr>
<td>2. APD=SLI= Dyslexia= ADHD</td>
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<td>3. SLI/ADHD/ Dyslexia LiD</td>
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<td>4. APD Language, Attention</td>
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<td>5. General Neurodevelopmental Disorder</td>
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**Auditory difficulties** |
**Language difficulties** |
**Attention difficulties** |
**Reading difficulties**

### Controversies in auditory processing disorder
The diagnostic term "APD" is used to address listening difficulties that somehow relate to deficits in the bottom-up and/or top-down processing of auditory information. Various professional audiology societies (e.g., ASHA, 1996; 2005 & American Academy of Audiology (AAA), 2010) stated in their APD guidance documents that APD is a problem of the central auditory system, separated from multi-modal cognitive and language problems. In these guidelines, APD is regarded as a consequence of a disturbed bottom-up function of the auditory system (Moore, 2012). According to Cacace and McFarland (2005; 2006), APD can only be seen as bottom-up deficit when there is a modality-specific disorder and the deficit only occurs in the processing of acoustic information and not when similar information is offered to other sensory modalities (for example the visual modality). Without modality specificity, the concept of APD has little power and significance (McFarland & Cacace, 2009). In contrast to the American audiology societies, there is, according to the British Society of Audiology (BSA, 2011), no evidence for the claim that APD is primarily a disturbed bottom-up processing. Research by Moore and colleagues (2010; 2011) have shown that poor listening in children in most cases has a cognitive component and that the listening problems are related to weak attention, poor working memory or language problems.
The lack of clarity about the nature of listening difficulties and controversies related to listening difficulties and APD, causes confusion at the theoretical level as well as at the clinical level. It is not clear how children with listening difficulties should be detected, diagnosed and treated. The problems of children with listening difficulties are diverse, with large inter-individual variation. No specific profile for a child with listening difficulties presently exists (Sharma, Purdy & Kelly, 2009; Ferguson et al., 2011; Miller & Wagstaff, 2011). The symptoms of children with reported listening difficulties and the association of these symptoms with other neurodevelopmental disorders are factors that influence the discussion about the validity of the APD diagnosis in addition to the discussion whether APD should be considered as a pure auditory bottom-up processing disorder or should be considered as a cognitive impairment.

Because of the lack of clarity and questions from clinicians about how to manage children who suffer from listening problems, these group of children are not always referred to the right discipline or multidisciplinary center. It is evident that a clinical demand exists for scientific insight into APD and that there is currently no clear, scientifically and evidence based diagnostic procedure for children with listening difficulties (Moore, Halliday & Amity, 2008).

**Audiology services in the Netherlands**

In the Netherlands, children with unexplained listening difficulties are usually identified by an education specialist and/or a speech-language therapist and after detection, referred by a general practitioner to an audiological center (Neijenhuis & Nijland, 2005; Neijenhuis & Van Herel-De Frel, 2010; Van den Bosch & Gerrits, 2013). An audiological center is an expertise center for hearing, speech and language. In an audiological center, professionals from different fields (e.g., audiologist, speech-language therapist, psychologist, child psychologist, linguist, social worker) work in multidisciplinary teams on diagnostics, rehabilitation and assistance of children and adults with an auditory and/or communicative disability (www.fenac.nl; www.audiologieboek.nl).

In response to questions from Dutch speech-language therapists working in private practices about uncertainties surrounding the signaling, referral and treatment of children with listening difficulties, the applied research project “Logopedic approach of auditory processing difficulties”, initiated by the Hanze University of applied sciences Groningen and funded by the Regional Attention and Action for Knowledge circulation (RAAK) of the Ministry of Education, Culture and Science in the Netherlands, was started in 2010. The aim of this project was, -1- to establish a uniform definition for the target group of children with listening difficulties, -2- to combine knowledge and experience of clinicians in the field of signaling, diagnosis, referral and treatment of children with listening difficulties and, -3- to make this knowledge accessible for professionals.

As part of this project, bachelor’s students from the Department of Speech and language Pathology at Hanze University of applied sciences Groningen studied the procedure of Dutch audiological centers with regard to referral, diagnostics and the follow-up program for children with unexplained listening difficulties. Based on the interviews with various professionals from Dutch audiological centers throughout the Netherlands, the conclusion could be made that there is a large variation between the Dutch centers in diagnostic procedures and counselling of children who experience listening difficulties (de Boer & Kuijpers, 2011). Since the first decade of this century, various auditory processing test batteries are available for the Dutch ACs (Neijenhuis & Van Herel-De Frel, 2010). It became clear that some centers used specific procedures for children who were referred with reported listening difficulties, including the use of the auditory processing test batteries, while other centers did not use auditory processing test batteries, but based their assessment on the broad mapping of the child’s functioning.
This diversity among Dutch audiological centers was two years later confirmed in a study of Van den Bosch and Gerrits (2013). They used a survey among Dutch speech-language therapists and audiologists, which showed that there was no uniformity in procedures for diagnostics and referral in the Netherlands for children with reduced listening ability.

The lack of uniformity in the Netherlands is in line with the (inter)national discussion about the definition, diagnostic route and the management of children with listening difficulties. There is quite some controversy surrounding the concepts of listening difficulties and APD (e.g., Chermak & Musiek, 1992; Cacace & McFarland, 1998; Moore, 2006; Cacace & McFarland, 2009; Fey et al., 2011; Moore et al., 2013). The definition, etiology, signs, symptoms, treatment and outcomes of difficulties in auditory processing have been discussed for more than 50 years already, and still there is little agreement among scientists and clinicians (Levy & Parkin, 2003; Hind, 2006; Moore, 2006; Cacace & McFarland, 2009; Beck, Clarke & Moore, 2016). The lack of clarity surrounding the referral pathway for children with listening difficulties appears to be caused by ambiguity about what difficulties in auditory processing are and the unknown cause of the listening difficulties in children (Hind, 2006). According to DeBonis and Moncrieff (2008) speech-language therapists are affected by this current state of uncertainty because their professional responsibilities includes screening for listening difficulties, making appropriate referrals, and providing intervention services.

**Aim of this thesis**

In conclusion, there is an urgent and pressing need for clarity about the etiology, definition, symptoms, diagnostics and management of children suffering from listening difficulties. This will advance the treatment of a vulnerable group of children. The overall aim of this PhD research is to investigate which behavioral characteristics are associated with listening difficulties and APD, how the relationship between APD and other neurodevelopmental disorders is, what the role of top-down processes is in children with listening difficulties, and to achieve, in collaboration with Dutch speech-language therapists and audiologists, a uniform and workable definition and working method for children with reported listening difficulties in the Netherlands.

**OUTLINE OF THIS THESIS**

The current PhD research has been carried out at the Hanze University of Applied Sciences (Hanze UAS) in collaboration with the Department of Otorhinolaryngology, Head & Neck Surgery at the University Medical Center Groningen (UMCG) and the Behavioural Science Institute at the Radboud University Nijmegen in the Netherlands. The research at the Hanze UAS is practice-based research and rooted in professional practice. The research aims to generate knowledge, insights and products that help solve the problems in professional practice and/or further development of this professional practice (Hanze UAS, 2017).

The central question of this thesis is whether APD is a truly disorder in auditory processing which can be regarded as a distinct and unique construct that can be distinguished theoretically and clinically from other neurodevelopmental disorders, such as SLI, dyslexia, ADHD and ASD. The following main question was formulated for this thesis:

- What are the characteristics of children with listening difficulties (LiD) and can auditory processing disorder (APD) regarded as a distinct clinical and identifiable disorder that explains the listening difficulties?
With regard to this main question, the following sub-questions were formulated:

1. Which characteristics are associated with (suspected) APD in children?
2. Which characteristics of children with (suspected) APD overlap with the characteristics of children diagnosed with another neurodevelopmental disorder, such as specific language impairment (SLI), dyslexia, attention deficit hyperactivity disorder (ADHD), and autism spectrum disorder (ASD)?
3. Are listening difficulties in children aged 8 to 12 related to auditory and/or visual attention skills?
4. What is a useful definition and working method for Dutch professionals who work with children with listening difficulties in clinical practice and how and by whom should listening difficulties in children be identified, examined and treated?

Part I
In the first part of the thesis, the fundamentals of listening difficulties and APD were studied. Since publications about APD often contain a series of personal opinions or clinical anecdotes that do not have a solid basis in theory or evidence, two systematic reviews were carried out to synthesize the existing knowledge about the characteristics of children diagnosed with APD and children with a suspicion of APD.

In Chapter 2, we systematically studied in the existing literature how children with an APD diagnosis perform on various tests and what possible characteristics are of APD. In addition, Chapter 3 presents our response to the letter to the editor of Moncrieff (2017): “Response to de Wit et al., 2016, Characteristics of Auditory Processing Disorders: A Systematic Review”. As described above, APD is a highly controversial subject in which a long-standing intellectual, theoretical and practical impasse exists. It is therefore not surprising that there is a lot of reaction to each other’s publications in scientific journals.

In Chapter 4, we systematically studied in the existing literature on how children with (suspected) APD perform various tests compared to children diagnosed with another developmental disorder. In this study, the overlap of the characteristics of children with (suspected) APD and children diagnosed with another neurodevelopmental disorder is described.

The underlying etiology and cause of listening difficulties in children is unclear. Recent evidence suggests that top-down processes, like attention, memory and language skills have a substantial impact on the listening skills of children. This is why we examined in Chapter 5 in an explorative study the differences in performances between children with reported listening difficulties and typically developing children on tests of communication, auditory processing, nonverbal intelligence, working memory, and visual and auditory attention.

Part II
In the second part of the thesis, the development and realization of a Dutch position statement for the professional practice is described. In practice-based research, the opinion of professionals from the field is an important factor to include besides scientific underpinnings.

In Chapter 6, a qualitative study is described in which the perspectives of various professionals from Dutch audiological centers on the definition and care pathways of children with suspected APD were studied with focus group discussions.

In Chapter 7, a two-round internet-based Delphi study is presented, which was used to reach consensus among a small group of speech-language therapists and audiologists from the clinical field on clinical signs, comorbidity and referral of children with listening difficulties.
In **Chapter 8**, the acquired knowledge is returned to the professional practice through the ‘Dutch Position Statement Children with Listening Difficulties’. At the time of the development of the Dutch Position Statement the information from chapter 2, 4, 6 and 7 was available. Therefore, the knowledge and evidence from these studies is included in the Dutch Position Statement. The purpose of this Position Statement is to provide professionals with tools to identify, diagnose and treat children with listening difficulties.

Finally, **Chapter 9**, summarizes and discusses the main outcomes of the studies in this thesis and the implications and future perspectives for children with listening difficulties and the professionals in the field.
REFERENCES


PART 1