Random Gap Detection Test and Random Gap Detection Test-Expanded results in children with auditory neuropathy

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1. Introduction

Auditory neuropathy is a term presently used to describe a condition, found in some patients ranging in age from infants to adults, in which the patient displays auditory characteristics consistent with normal outer hair cell function and abnormal
neural function at the level of the VIIIth (vestibulo–cochlear) nerve. These characteristics are observed on clinical audiologic tests as normal otoacoustic emissions (OAEs) in the presence of an absent or severely abnormal auditory brainstem response (ABR) [1]. Patients with auditory neuropathy/dysssynchrony exhibit no auditory brainstem response (ABR), no middle ear muscle response, and both normal otoacoustic emissions or normal cochlear microphonics [2]. These patients are distinguished from patients with space-occupying lesions, such as VIIIth nerve tumors, or multiple sclerosis, in that radiological evaluation yields normal results and even the most peripheral responses from the VIIIth nerve are absent. Patients with auditory neuropathy require a different management approach to their auditory and communication problems from approaches used with patients with usual peripheral hearing losses [1].

Auditory neuropathy affects the normal synchronous activity in the auditory nerve, without affecting the amplification function in the inner ear. Patients with auditory neuropathy often complain that they can hear sounds, but cannot understand speech. Starr et al. [3] reported psychophysical tests indicating that these patients’ poor speech recognition is due to a severe impairment in their temporal processing abilities. In some patients, this difficulty may be related to neural timing problems that may limit the ability to follow rapid transitions of normal speech.

The patients with auditory neuropathy display some awareness of sound around them. They do not have the advantage of accurate auditory information to help them discriminate and learn appropriate speech and language patterns. Since speech and language develop largely through repetition of heard patterns. Until the underlying etiologies of auditory neuropathy are better understood, the appropriateness of using hearing aids and cochlear implants is difficult to determine. Hearing aids are being tried to a limited extent in some children with auditory neuropathy. If the clinician or a parent strongly wishes to try a hearing aid to enhance awareness of sound, then we recommend high quality, low gain, and wide dynamic range compression hearing aids. The potential benefit of cochlear implants is still an open question. If the underlying etiology of the auditory neuropathy in a particular patient is cochlear in origin (i.e., the inner hair cells and/or the hair cell–nerve juncture) and neural function is intact, then a cochlear implant may be potentially beneficial [4–6].

Rance et al. [7] found perceptual characterization of children with auditory neuropathy. Frequency resolution (notched noise masking) results for the AN subjects were equivalent to those of the normal-hearing subjects reflecting the “normal” outer hair cell function that characterizes the AN condition. Temporal resolution (TMTF) findings were, however, abnormal in many AN subjects and the degree of temporal disruption was correlated with speech discrimination (CNC) score. Frequency discrimination ability (for both fixed and frequency modulated stimuli) was also affected in those children with poor temporal resolution [7]. The temporal resolution of the auditory system is exquisite, with neural systems capable of submillisecond resolution in decoding features in the acoustic signal [8].

Auditory neuropathy is a hearing disorder in which sound enters the inner ear normally but the transmission of signals from the inner ear to the brain is impaired [9]. AN is quite different from the sensorineural hearing loss (SNHL). If the patients have only SNHL, they can manage to perform speech perception by amplification. When hearing is provided by hearing aids or cochlear implants, speech perception may be improved. Whereas, in AN patients, there are not only SNHL but also extremely temporal processing disorders. Speech perception was abnormal. Their speech perceptions cannot be elevated by amplification (hearing aids or cochlear implants). This difference can be seen in Most and Peled’s study [10]. They assessed perception of suprasegmental features of speech by 30 prelingual children with sensorineural hearing loss. Ten children had cochlear implants (CIs), and 20 children wore hearing aids (HA): 10 with severe hearing loss and 10 with profound hearing loss. Perception of intonation, syllable stress, word emphasis, and word pattern was assessed. Results revealed that the two HA groups significantly outperformed the CI group in perceiving both intonation and stress. Within each group, word pattern was perceived best, and then intonation and emphasis, with syllable stress perceived poorest. No significant correlation emerged between age at implantation and perception of the various suprasegmental features, possibly due to participants’ relatively late age at implantation [10]. Results indicated that there was no difference between these children with hearing loss and auditory neuropathy. In hearing loss, after the amplification by hearing aids or cochlear implants, speech perception and gap detection were normal. But, in AN group, amplification by hearing aids or cochlear implantation is not enough. There are also temporal processing disorders, therefore speech perception and gap detection results were not normal.

Zeng et al. [11] studied to answer the following two questions: does noise present a particular problem for people with AN? Can clear speech and cochlear implants alleviate this problem? They studied 13 participants with AN. Of these participants, 7 had received a cochlear implant. Eight sentence-recognition experiments were conducted to examine the clear speech advantage in two listening conditions (quiet and noise) using four stimulation modes (monaural acoustic, diotic acoustic, monaural electric and binaurally combined acoustic and electric stimulation). Participants with AN most likely derive the clear speech advantage from enhanced temporal properties in clear speech and improved neural synchrony with electric stimulation. Although the present result supports cochlear implantation as one treatment choice for people with AN, it suggests that the use of innovative hearing aids may be another viable option to improve speech perception in noise.

In Zilotti and Desgualdo’s study [12], the RGDT was applied to 236 subjects ranging from 5 to 56 years of age. All subjects had normal hearing and middle ear function. One hundred subjects in group one had normal auditory processing while 136 subjects in group two had auditory processing disorders. Groups were further subdivided according to age (5–6 years; 7–8 years; 9–11 years; 12–25 years; and 26–53 years). Results of the study found a statistically significant difference between groups (0.001) with the mean gap detection threshold in group one of 6.7 ms and group two of 32.1 ms. No significant differences were observed when considering age range for each frequency [12].

In auditory neuropathy (AN) children with hearing aids (HAs) or cochlear implant (CI) [3–5], the speech perception improvement may not be as expected. These children may perform speech perception after a few repeats. This condition may show that these children had difficulties in receiving and processing speech sounds. If the children with AN cannot distinguish the heard tones one or two in Random Gap Detection Test (RGDT), a test of temporal processing (auditory timing) ability [13]; their benefit performances between hearing aids or CI may not be significant. It is thought that the answer of this question is closely related with unique auditory processing performance of each child. Therefore, we planned this study which aimed to investigate the RGDT and RGDT-Expanded (RGDT-EXP) performance of five children with AN compared to normal hearing children; and the effects of AN on auditory timing function. In the literature, we could not find any similar studies on this matter.

2. Materials and methods

The study was assessed in Hacettepe University, Faculty of Medicine, Division of Audiology and Speech Pathology of ENT
Department. All steps of the study were planned and continued according to the principles outlined in the Declaration of Helsinki [14].

2.1. Subjects

The patients with auditory neuropathy according to diagnosis criteria [1,15] were sent from Hacettepe University, Faculty of Medicine ENT Department, Division of Audiology.

Diagnosis of AN was performed with the criteria of [1,15]:

1. In pure tone thresholds, there was sensorineural hearing loss and/or fluctuant hearing loss;
2. otocoustic emissions: normal;
3. middle ear muscle reflexes: ipsilateral: absent, contralateral: absent;
4. in ABR: ABR waves were absent (or severely abnormal); and cochlear microphonics were absent.

The study group was consisted of five children (3 male, 2 female) with auditory neuropathy (AN) between ages of 7 and 13 years. The control group was consisted of 10 normal hearing children who had not auditory processing problem between ages of 7 and 16 years (5 male, 5 female).

In AN group, hearing loss was prelingual in one child and postlingual in four children. History of children with auditory neuropathy (AN) was shown in Table 1. The knowledge about children’s story including etiological factors of hearing losses and specific auditory behaviors was taken from parents for small children and from child’s own for bigger children as written. All children in the study and control groups were included into the study with their parents’ agreement by written informed consent to participate the study, and to give permission for the use of their child’s all data.

In this study, RGDT and RGDT-EXP [13] were applied to five children with auditory neuropathy and 10 children of the control group. In the first test, all children were applied to RGDT and RGDT-EXP. Each child responded whether he/she heard one or two tones. Their responses were taken as verbally and/or hold up one finger or two fingers. In the second test, they were applied speech discrimination test in quiet environment and in noise. Normal gap detection threshold is considered to be between 2 and 20 ms [13].

2.2. Procedure

2.2.1. Random Gap Detection Test (RGDT) [13]

The RGDT is a test of temporal processing (auditory timing) ability. Disorders of auditory timing are related to disorders of auditory discrimination, reading and language. Stimulus pairs with 0–40 ms gaps are presented. Individual identifies when one or two tones are heard. Normal gap detection occurs at 20 ms or less.

The auditory gap detection threshold of tones and white noise (clicks) is obtained by having the subject identify when signal pairs are separated in time from 0 to 40 ms. The major improvement in the signal presentation during the RGDT is that the gap interval is randomly assigned, and therefore unpredictable to the subject. The test includes stimuli at four frequencies (500, 1000, 2000, and 4000 Hz) and white noise clicks of 50 μs duration. A practice session is presented with tone pairs at 1000 Hz.

2.2.2. Description of the stimuli

Each stimulus is composed of a pair of tone pulses. Each pulse has duration of 17 ms, including a 1 ms rise–fall time. The silent intervals between the two pulses varies from 50 to 300 ms. The test includes four frequencies: 500, 100, 2000, and 4000 Hz.

2.2.3. Scores reported

- Gap detection thresholds (GDTs) are reported for each frequency tested: 500, 100, 2000, and 4000 Hz. Identified lowest gap is detected for each of the frequencies 500–4000 Hz.
- The composite gap detection threshold (CGDT) is the average of results reported across the four test frequencies of 500–4000 Hz.

Normal GDT is considered to be between 2 and 20 ms [13]. When a subject fails the RGDT and gap detection threshold exceed 50 ms the RGDT-Expanded is administered. Test results indicated that any subject with RGDT thresholds greater than 20 ms is likely to have temporal processing deficits that interfere with normal speech perception, and phoneme recognition [13].

2.2.4. The RGDT-Expanded test (RGDT-EXP) [13]

RGDT-EXP is intended for individuals whose gap detection threshold exceeds 50 ms. This test begins at time intervals longer than those measured by the standard RGDT, and includes time intervals between 50 and 300 ms. The test is administered in the same manner as the standard RGDT. Individuals who require this test to establish a gap detection threshold have already demonstrated abnormal temporal processing abilities. The single purpose is to determine the time interval in which their gap detection thresholds exist.

2.2.5. Administration procedures

The stimuli presented through an audiometer, earphones connected directly to CD player, presented the signal binaurally at the comfortable listening level according to the child (55 dB between 65 dB). The test takes approximately 10 min to administer, including instructions and practice. Interpretation is made by averaging the gap detection threshold for all tonal stimuli and comparing the results to normative data that is currently available for children over 5 years old.

2.2.6. Child’s response

All of children had to respond verbally or hold up one finger or two fingers whether he/she heard one or two tones. In healthy

Table 1

<table>
<thead>
<tr>
<th>Age and gender</th>
<th>Etiology</th>
<th>Onset age of hearing loss</th>
<th>Diagnosis</th>
<th>Hearing aid usage</th>
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</thead>
<tbody>
<tr>
<td>Case 1 female (16 years 2 months)</td>
<td>Sudden idiopathic hearing lost</td>
<td>8 years</td>
<td>Mild to moderate HL</td>
<td>She cannot use hearing aid</td>
</tr>
<tr>
<td>Case 2 Female (9 years 11 months)</td>
<td>Sudden idiopathic hearing lost</td>
<td>5 years 6 months</td>
<td>Moderate to severe HL</td>
<td>Occasionally</td>
</tr>
<tr>
<td>Case 3 male (7 years 3 months)</td>
<td>Prelingual AN. Etiology unknown</td>
<td>Unknown</td>
<td>Mild to moderate HL</td>
<td>Occasionally</td>
</tr>
<tr>
<td>Case 4 male (10 years 9 months)</td>
<td>Sudden idiopathic hearing lost</td>
<td>9 years</td>
<td>Moderate to severe HL</td>
<td>He has hearing aid</td>
</tr>
<tr>
<td>Case 5 male (13 years 8 months)</td>
<td>Sudden idiopathic hearing lost</td>
<td>5 years</td>
<td>Severe HL</td>
<td>He used hearing aid for 1 year. Now, he has CI (right ear)</td>
</tr>
</tbody>
</table>

* Case 4 and Case 5 are brothers.
children without hearing loss and without AN, normal value of gap detection threshold was <20 ms [13].

2.3. Statistical analysis

Statistical packet for SPSS (Version 9.0) was used for statistical evaluation.

At each frequency (500–4000 Hz) of RGDT, the difference between GDTs and CGDTs was analyzed by "Mann–Whitney U test". P value < 0.05 was considered as statistically significant.

3. Results

History of children with AN was demonstrated in Table 1. Minimum detectable gap (gap detection threshold) with RGDT/RGDT-EXP Tests at 500–4000 Hz; and composite gap detection thresholds for each child of the control and study (AN) groups were shown in Table 2. Any of the children with AN could not be able to perform RGDT. Therefore the RGDT-EXP was applied in this group. In the study group, GDTs were all over 50 ms for all children included into the study group with AN, mean of the GDTs was all over the normal limits; and in control group, mean of GDTs was all in normal limits. The difference between the mean GDTs of the study group was significantly higher than the control groups at all frequencies of 500 Hz (P = 0.002), 1000 Hz (P = 0.002), 2000 Hz (P = 0.002) and 4000 Hz (P = 0.002) by Mann–Whitney U test.

In AN group, CGDT (97.5 ± 9.57 ms) was significantly higher than that of the control group (10.35 ± 0.65 ms).

4. Discussion

Auditory neuropathy/dysynchrony is a form of hearing impairment in which cochlear outer hair cell function is spared but neural transmission in the auditory pathway is disordered. A distinguishing characteristic of an AN/AD is the presence of auditory responses associated with normal function of the outer hair cells of the cochlea and poor neural synchrony of the auditory (VIIIth cranial) nerve [16].

Zeng and Liu [17] conducted two sets of experimental experiments to test the relationship between temporal (processing of acoustic stimuli over time: understand speech in quiet and in background noise) and speech processing deficits in auditory neuropathy. In the first set of experiments, psychophysical data were collected in temporal integration, gap detection, and temporal modulation transfer function in auditory neuropathy subjects and three groups of control subjects. In the second set of experiments, acoustic simulations were developed based on the measured psychophysical data in auditory neuropathy subjects and were validated by obtaining similarly impaired temporal and speech processing.

Table 2

<table>
<thead>
<tr>
<th>Frequencies</th>
<th>Groups</th>
<th>Mean (ms)</th>
<th>Standard deviation</th>
<th>Minimum</th>
<th>Maximum</th>
</tr>
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<td>500 Hz</td>
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<td>89.44</td>
<td>50.00</td>
<td>250.00</td>
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<tr>
<td>1000 Hz</td>
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<td>2000 Hz</td>
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<td>150.00</td>
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<tr>
<td>4000 Hz</td>
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<tr>
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<td>9.57</td>
<td>90.00</td>
<td>110.00</td>
</tr>
<tr>
<td>500 Hz</td>
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<td>10.12</td>
<td>0.00</td>
<td>25.00</td>
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<tr>
<td>1000 Hz</td>
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<td>20.00</td>
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<td>9.05</td>
<td>0.00</td>
<td>25.00</td>
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<tr>
<td>4000 Hz</td>
<td>Control group</td>
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<td>8.81</td>
<td>0.00</td>
<td>25.00</td>
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<tr>
<td>CGDT</td>
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P value shows the results of Mann–Whitney U test.
results in normally hearing subjects. They studied eight patients with auditory neuropathy, including one with unilateral neuropathy. They also studied three controls, including the healthy ear in the unilateral neuropathy subject, one cochlear-impaired subject with a low-frequency hearing loss and six normally hearing subjects. The cochlear-impaired subject was chosen because of his unusual configuration of hearing loss. They found that temporal processing abilities were severely impaired in auditory neuropathy subjects. They said that; while the exact physiological process underlying auditory neuropathy is not clear, the temporal processing deficit may be a result of desynchronized neural activity at the auditory nerve level.

Michalewski et al. [18] evaluated auditory temporal processing in a group of normal-hearing subjects and in a group of hearing-impaired individuals with auditory neuropathy (AN) using electrophysiological and psychoacoustic methods. They found a close association between gap detection thresholds measured psychoacoustically and electrophysiologically in both normals and in AN subjects.

In the present study, any of the children with AN could not be able to perform RGDT. Therefore the RGDT-EXP was applied in this group. In the study group, GDTs were all over 50 ms at 500–4000 Hz; and CGDTs were all over 50 ms for all children included into the study group with AN. In control group, except child 9 (GDTs were 25 ms at 3000 and 4000 Hz); and child 10 (GDT was 25 ms at 500 Hz); GDTs were all normal limits for 500–4000 Hz for all children included into the study group. CGDTs were all in normal limits for the control group, except child 9 (CGDT were 22.50, slightly higher than normal limits).

In the study group with AN, mean of the GDTs was all over the normal limits; and in control group, mean of GDTs was all in normal limits. The difference between the mean GDTs of the study group was significantly higher than the control groups at all frequencies of 500–4000. In AN group, CGDT (97.5 ± 9.57 ms) was significantly higher than that of the control group (10.35 ± 0.65 ms).

In the present study, our results have shown that children with AN who have hearing aids and cochlear implant; or no hearing aids; cannot differentiate the gap detection threshold in optimal time compared with controls. Such as, Case 4 and Case 5. They were two brothers and cannot find any difference between the gap detection threshold by RGDT-EXP (mean: 50 ms). In Case 5 who has a CI and AN, could not make any speech discrimination, because he has a very prolonged gap detection threshold than normal. If children cannot detect or discriminate the order of two tones at rates of 20 ms, the children will be unable to perceive rapid changes in formant frequencies of ongoing speech. Therefore, they are unable to perceive rapid changes in formant frequencies of ongoing speech. This finding also agrees with their daily life. In our study, we observed that these children can understand the speech only with only several repeats in their daily life. Because, their gap detections were much worse for short duration stimuli than for longer stimuli.

This study quantifies disorders of timing in the auditory system (called temporal processing disorders) in children with AN. Our study is quite important for the literature, even if limited cases of AN in the study group. Because, our findings will help to show why the children with AN cannot understand the speech sounds they heard. It was obviously seen that the children with AN cannot differentiate the gap detection threshold in ongoing speech. Such as, rapidly changing sounds such as /ɪ/, /i/. Temporal processing disorders are related to phonologic processing deficits, problems of auditory discrimination, and receptive language. Listening (audibility of acoustic feature) the smaller gap that can be detected, such as, /s/, /f/ may be inaudible in a noisy environment. If a child with auditory neuropathy cannot detect or discriminate the order of two tones at rates at least as fast as 20 ms, the child will be unable to correctly perceive ongoing speech. Thus, deficits in “hearing” small differences in timing aspect of ongoing speech create speech discrimination errors, even though hearing thresholds may be normal [13].

In our study, children with AN differentiate whether the heard tones were one or two in very long time over the normal limits by RGDT-EXP. These results may show that the children with AN may not benefit from hearing aid or CI in terms of gap differentiation as expected. However, Tomblin et al. [19] reported the comparison of language achievement in children with cochlear implants and children using hearing aids. They found clear differences between with and without CI experience of 2 years of post-implant period. It was found that CI users achieved significantly better scores. Blamey et al. [20] evaluated 87 primary-school children with impaired hearing. They evaluated using speech perception, production, and language measures over a 3-year period. Forty-seven children with a mean unaided pure tone-average hearing loss of 106 dB HL used a 22-electrode cochlear implant, and 40 with a mean unaided pure tone-average hearing loss of 78 dB HL were fitted with hearing aids. Rates of improvement for individual children were not correlated significantly with the degree of hearing loss. Their language and speech perception scores in the auditory test condition showed a slight downward trend over time.

Rance and Barker [21] evaluated speech perception skills in children with auditory neuropathy (AN)/auditory dysynchrony (AD)-type hearing loss managed with either hearing aids or cochlear implants. Prospective data collection in three subject groups: AN/AD children fitted with bilateral amplification, AN/AD children fitted with cochlear implant (in one or both ears), and a matched control group of implanted children with sensorineural hearing loss. In the results of this study, of the 10 implanted AN/AD children, 9 demonstrated significant speech discrimination (consonant-nucleus-consonant phoneme score ≥ 55%). Similar results were obtained for the aided AN/AD group. Findings for both AN/AD subject groups were poorer than those of the implanted sensorineural cohort. They concluded that cochlear implantation can offer useful hearing in subjects with AN/AD-type hearing loss. However, expectations for this group may need to be lower than for patients with peripheral (cochlear) loss.

In our clinic, auditory training was given to these children; but no meaningful recovery was observed. As it is known, the children with AN cannot distinguish acoustic sounds and speech sounds; or may hear as incorrect, they cannot understand what they heard. If
they distinguish the sounds of language, in that time, there will be no detected learning difficulties of language and speech anymore.

Finally, auditory neuropathy is defined as the disorder of VIIIth cranial nerve synchronization. The RGDT is viewed as a test of temporal integrity at the level of the cortex [13]. In the present study, children with AN cannot perform gap detection in normal time limits; and cause of the problem may not only be at VIIIth cranial nerve level. These results may indicate processing disorder at the cortical level. Because, test results indicated that any subject with RGDT thresholds greater that 20 ms is likely to have temporal processing deficits that interfere with normal speech perception, and phoneme recognition.

According to these results of gap detections; and because of this dilemma, future CI and hearing aid processors need to extract and encode these acoustic cues to achieve better performance in tone perception and production. In addition, perceptual training may address temporal aspects of audition.

Conflict of interest

The authors declare that there is no conflict of interest.

References