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# **Research Paper**



# Is tinnitus a major concern in individuals with auditory neuropathy spectrum disorder? – Questionnaire based study

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KEYWORDS Tinnitus; Functional impairment; Severity; Questionnaire	Abstract Objective: Our study attempted to determine if tinnitus is a major concern in individuals with auditory neuropathy spectrum disorder (ANSD). The study also attempted to determine if pure tone average (PTA), speech identification scores (SIS), or reported duration of hearing loss could predict the tinnitus severity and functional handicap in adults with ANSD. <i>Methods:</i> Tinnitus functional index (TFI) and tinnitus severity index (TSI) were administered on all the 40 participants diagnosed with ANSD. <i>Results:</i> The mean TFI score was 44.3, which suggested moderate degree of tinnitus related functional impairment. The mean TSI score was found to be 33.3, which suggested moderate degree of severity in individuals with ANSD. The study also showed that tinnitus causes functional impairment that leading to sleep disturbances, emotional problems, and affecting the quality of life. There was no significant correlation ( $P > 0.05$ ) between PTA, SIS, reported onset of hearing loss and scores of TFI or TSI. <i>Conclusion:</i> Tinnitus in individuals with ANSD should also be given priority during assessment and provide appropriate tinnitus management. Copyright © 2017 Chinese Medical Association. Production and hosting by Elsevier B.V. on behalf of KeAi Communications Co., Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
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Auditory neuropathy spectrum disorder (ANSD) can be defined as a clinical disorder, in which clients exhibit normal otoacoustic emissions (OAE), but abnormal or absent auditory brainstem response (ABR).<sup>1–6</sup> The prevalence rates of ANSD in western countries are reported to vary from 11% to 0.5%.<sup>7–12</sup> In Indian population, Kumar and Jayaram<sup>13</sup> reported that 1 in 183 were diagnosed as ANSD among individuals with sensorineural hearing loss. The reviews of literature suggested that majority of the individuals with ANSD suffered reduced hearing sensitivity and difficulty in understanding speech. Besides, tinnitus was also found to be one of the complaints.<sup>5,13,14</sup>

Chandan et al<sup>14</sup> reported that tinnitus was found in 63.75% of individuals with ANSD (95 out of 149). Prabhu et al<sup>5</sup> reported that tinnitus was reported in 83.3% of the participants (25 out of 30) in their study. Kumar and Jayaram<sup>13</sup> also reported that bilateral tinnitus is present in 50% of the individuals with ANSD. Prabhu and Sneha<sup>15</sup> administered self report tinnitus handicap questionnaire (SR-THQ) on 30 individuals with ANSD. They found that majority of individuals with ANSD showed moderate degree of tinnitus handicap. In addition, the tinnitus could also cause significant emotional problems and be catastrophic in few individuals with ANSD based on this guestionnaire study. Thus, previous literature suggested that tinnitus is one of the common symptoms affecting the individuals with ANSD. However, there is a dearth of literature that attempted to determine the severity of tinnitus and its impact on daily life functioning. In addition, none of the studies correlated audiological findings with scores obtained on tinnitus questionnaires.

To overcome these drawbacks, this study attempts to determine if tinnitus is a serious concern that affecting ANSD patients with different guestionnaires. Tinnitus functional index (TFI) and tinnitus severity index (TSI) were used to determine the degree and nature of tinnitus related problems experienced by individuals with ANSD. TFI developed by Meikle et al<sup>16</sup> consists of 25 guestions with eight sub-scales. The sub-scales of TFI include intrusive (unpleasantness, intrusiveness, and persistence), sense of control (reduced sense of control), cognitive (cognitive interference), sleep (sleep disturbance), auditory (auditory difficulties attributed to tinnitus), relaxation (interference with relaxation), guality of life (reduced quality of life), and emotional (emotional distress). TSI was developed by Meikle et al<sup>17</sup> that consists of 12 questions with 5-point rating scale. The severity of tinnitus can be classified into very mild category,<sup>1-12</sup> mild,<sup>13-24</sup> moderate (25-36), severe (37-48), and catastrophic (49-60) based on TSI scores. Thus, the aim of the study was to determine the severity of tinnitus and its effects on daily life functioning using TFI and TSI in ANSD individuals. In addition, it was also attempted to determine if pure tone average (PTA), speech identification scores (SIS), or reported duration of hearing loss could predict the tinnitus severity and functional handicap in adults with ANSD.

#### Material and methods

#### Participants

A total of 40 adults with ANSD with a complaint of bilateral tinnitus were considered for the study. The study sample consisted of 18 males and 22 females between the age ranges of 14–35 years (mean = 21.3 yrs, SD = 9.29). All the participants had pure tone average thresholds (PTA, average pure tone thresholds at 500 Hz, 1 kHz, 2 kHz and 4 kHz) ranging from mild (26-40 dB HL), moderate (41-55 dB HL), moderately severe (56-70 dB HL) to severe (71-90 dB HL) degree of hearing loss with 10 participants in each group.<sup>18</sup> There speech recognition thresholds (SRT) were poorer than PTA (>15 dB) and the speech identification scores (SIS) were poor (<60%) in all the participants of the study. Psychoacoustic evaluation of tinnitus was carried out on all the participants of the study. They were diagnosed as having ANSD based on presence of transient evoked otoacoustic emissions (TEOAE) and distortion product otoacoustic emissions (DPOAE), but absent of auditory brainstem responses. In addition, all the participants had no history nor presence of middle ear pathology with A type tympanogram,<sup>19</sup> in addition to missing acoustic reflexes. The criteria adopted to diagnose ANSD in the audiology clinic were recommended by Starr et al.<sup>20</sup> The diagnoses of ANSD were further confirmed by a neurologist. The duration of reported duration of hearing loss ranged from 12 months to 180 months. Hearing aids were not beneficial in all the participants of the study.

#### Procedure

Pure tone air conduction (AC) and bone conduction (BC) thresholds were estimated using modified Hughson and Westlake procedure.<sup>21</sup> AC thresholds were obtained for pure tone frequencies from 250 Hz to 8 kHz and BC thresholds from 250 Hz to 4 kHz at octave frequencies. A two-channel diagnostic audiometer was used to obtain air conduction and bone conduction pure tone thresholds. together with speech identification scores. Speech identification scores were obtained using headphones for phonemically balanced words developed for adults in Kannada by Yathiraj and Vijayalakshmi.<sup>22</sup> Recorded word lists were routed from a personal computer through a twochannel diagnostic audiometer at 40 dB SL (re: SRT). Psychoacoustic evaluation of tinnitus determined according to the procedure described by Prabhu and Chandan.<sup>23</sup> The tinnitus pitch and loudness was estimated in all the participants.

An immittance meter GrasonStadler Inc. Tympstar (GSI-TS) was used for immittance testing. The better ear of the participant was tested to obtain tympanogram and acoustic reflexes for a probe tone frequency of 226 Hz. Acoustic reflexes were measured using 500, 1000, 2000, and 4000 Hz pure tones, which presented to both ipsi-lateral and contralateral ears. Otodynamics ILO v.6 OAE analyzer was used to obtain TEOAE and DPOAE. After ensuring adequate probe fit, TEOAE were measured for non-linear click trains presented at 80 dB peSPL. DPOAE were recorded with tones F1 and F2 at the ratio of 1.22, while at the intensities of 65 dB SPL and 55 dB SPL, respectively. If the overall signal to noise ratio was more than 6 dB for TEOAE and DPOAE respectively, with the waveform reproducibility was more than 50% at least at two frequency bands,<sup>24,25</sup> OAE would be considered as present. Biologic Navigator Pro (Bio-logic, Mundelein, IL) AEP system with ER 3A insert earphones was used to record ABR. Click evoked ABR was recorded twice and replicated for 100 µs click stimuli delivered at a repetition rate of 11.1 clicks/second at 90 dB nHL. The recordings were obtained for a total of 1500 sweeps while a filter setting of 100 Hz-3000 Hz was used. ABR was considered as absent if peaks were not clearly identified in both the recordings and lacked replications.

TFI was administered according the procedure suggested by Meikle et al.<sup>16</sup> The total TFI score and the scores obtained for all the eight sub-scales (intrusiveness, sense of control, cognitive interference, sleep disturbance, auditory issues, relaxation issues, quality of life, and emotional distress) were analyzed. Less than 25 in TFI score indicates mild tinnitus, which left little or no need of intervention. The score ranging from 25 to 50 means significant problem with tinnitus that intervention is required. A TFI score of greater than 50 is considered as severe tinnitus, who would need warrants aggressive interventions. TSI was administered according to the procedure suggested by Meikle et al.<sup>17</sup> It consists of 12 questions answered on a five-point rating scale with a total score of 60 points. Based on the score, the severity of tinnitus (very mild, mild, moderate, severe or catastrophic) was determined. Multiple regression analysis was also done to determine the predictors, like PTA, SIS, or reported onset of hearing loss of TSI and TFI scores.

#### Ethical considerations

In this study, all the testing procedures done were using non-invasive technique adhering to conditions of ethical approval committee of the institute. After explaining test procedure to the patients and their family members, informed consents were obtained from all the patients or their family members for participating in the study before testing.

### Results

Psychoacoustic evaluation of tinnitus results suggests that 80% of participants (32 out of 40) had low-pitched tinnitus (<1000 Hz). The intensity of tinnitus was approximately 10–15 dB in all this study. The range of TFI scores in the study was 18–70 indicates mild to severe tinnitus related functional difficulty. The mean TFI score was 44.3 (SD = 16.53) suggesting that majority of the participants had moderate degree of tinnitus related functional impairment. The mean scores for sub-scales of TFI were 21.8 (SD = 9.7) for intrusiveness, 37.6 (SD = 22.7) for sense of control, 22.2 (SD = 11.3) for cognitive interference, 63.5 (SD = 22.2) for sleep disturbance, 59.7 (SD = 17.1) for auditory issues, 33.2 (SD = 13.3) for relaxation issues, 56.6

(SD = 28.1) for quality of life, and 52.4 (SD = 19.9) for emotional distress. These results suggested that all the subscales were affected. Especially, sleep disturbances, auditory difficulties, emotional problems, and reduced quality of life scores were relatively higher. The mean and SD of scores across different sub-scales are shown in Fig. 1.

The results of TSI showed that the score ranged from 14 to 45 in this study with a mean TSI score of 33.3 (SD = 14.2), which suggested majority of participants with ANSD suffered a moderate tinnitus with a mild to severe degree of severity. Within the group, 10 out of 40 (25%) had mild degree, 22 out of 40 (55%) had moderate degree, and 8 out of 40 (20%) had severe degree of tinnitus as shown in Fig. 2.

Multiple regression analyses showed that none of the factors, PTA, SIS or reported onset of hearing loss, could predict the TFI and TSI scores. The regression model for TFI and these predictors showed  $R^2 = 0.13$  and F (3, 36) = 1.81, P > 0.05 suggesting a poor correlation. Similarly, regression model for TSI and the predictors showed  $R^2 = 0.08$  and F (3, 36) = 1.08, P > 0.05 again suggesting a very poor correlation. The details of multiple regression analyses are provided in Table 1.

#### Discussion

The results of the study showed that individuals with ANSD could exhibit a moderate degree of tinnitus related functional impairment, which leads to sleep disturbances, emotional problems, exacerbating auditory difficulties, and reduced guality of life. The functional difficulties faced by ANSD patients were usually attributed to poor speech perception and communication difficulties. Thus, the audiologists usually ignore the tinnitus in these patients and its possible impact on quality of life. This study, in contrast, suggested that tinnitus could also significantly affect the daily life functioning. It can be severe enough to cause emotional problems, sleep disturbances, and exacerbates auditory problems. Tinnitus can disrupt sleep and the ability to concentrate at work or in social interactions, which can cause depression, irritability, frustration, stress, and feelings of helplessness.<sup>26</sup> Previously, Prabhu and Sneha<sup>15</sup> also reported tinnitus caused functional challenges that were catastrophic in some ANSD individuals.

The results of the study also showed that majority of ANSD patients had moderate degree of tinnitus, the severity of which ranged from mild to severe. Meikle et al<sup>17</sup> suggested that if the severity of tinnitus were more than mild degree, it requires intervention. In this study, about 20% participants (8 out of 40) suffered severe tinnitus. Hence, appropriate tinnitus management strategies could to be applied according to present guideline. Thus, the results of this study pointed out the importance of tinnitus intervention in individuals with ANSD, which were usually ignored before. Additionally, this study also showed no correlation between the questionnaire scores and predictors, such as PTA, SIS, and reported onset of hearing loss. These results suggested that the tinnitus severity and functional handicap are independent to onset of hearing loss, degree of hearing loss, and speech perception abilities.

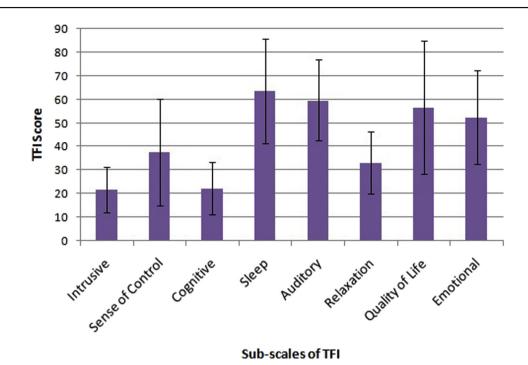


Fig. 1 The mean and standard deviation of scores obtained across eight different sub-scales of TFI.

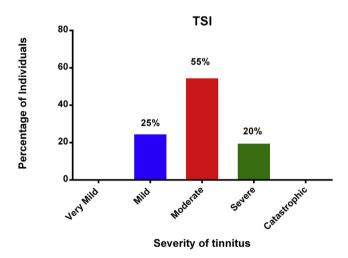


Fig. 2 The percentage of individuals across severity of tinnitus measured using TSI.

## Limitations of the study and future directions

The present study was carried out using English version of TFI and TSI, which is not standardized for Indian population. There is a need to repeat the study with standardized hearing handicap questionnaires in the local language of individuals with ANSD. In addition, considering the small sample size, results in general should be interpreted cautiously. Further studies with larger sample size are essential to confirm the results obtained in this study. The changes in tinnitus handicap score with tinnitus treatment in ANSD patients should also be addressed in future.

# Conclusion

This study attempted to determine if tinnitus is a major concern in individuals with ANSD. Tinnitus handicap questionnaire, TFI and TSI, were administered on all the participants in the study. The results showed that individuals with ANSD reported moderate degree of tinnitus related functional impairment. The severity of tinnitus was also moderate degree in majority of the participants. The study

Table 1Results of multiple regression analysis of TFI and TSI scores with duration of hearing loss, pure tone average (PTA) andspeech identification scores (SIS) as the predictors.

Independent variable	TFI			TSI		
	Pearson's correlation	β	R <sup>2</sup>	Pearson's Correlation	β	R <sup>2</sup>
SIS	0.21	0.25	0.13	0.83*	0.18	0.08
ΡΤΑ	0.17	0.19		-0.05	0.12	
Duration of hearing loss	-0.03	0.04		0.21	-0.19	

also showed that tinnitus causes functional impairment causing sleep disturbances, emotional problems, and reduced quality of life. There was no correlation between PTA, SIS, and reported onset of hearing loss to scores of two questionnaires. Thus, the present study suggests that symptom of tinnitus in individuals with ANSD should be paid attention during assessment along with other auditory symptoms and provided appropriate management.

# **Conflicts of interest**

The author reports no conflicts of interest. The author alone is responsible for the content and writing of the paper.

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