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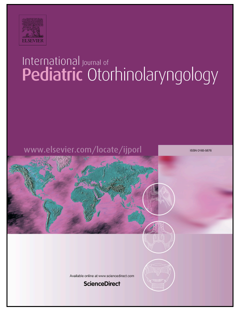

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Distinguishing between Hyperacusis and Misophonia in children with Auditory Processing Disorder (APD)

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ABSTRACT

Objectives: Decreased sound tolerance (DST) is common in children with auditory processing disorder (APD). This study aimed to differentiate between hyperacusis and misophonia in children with APD.

Design: A retrospective study evaluating outcomes of structured history and co-morbidity following Research Domain Criteria (RDoC) frame-work. Misophonia was considered as oversensitivity to eating/chewing sounds and hyperacusis as oversensitivity to other sounds.

Study sample: Two hundred and seventy-nine children (160 males;119 females), 6-16 year-olds with NVIQ ≥80, diagnosed with APD between January 2021 and December 2022.

Results: One hundred and forty-three out of 279 children with APD had DST, of which 107 had hyperacusis (without misophonia) and 36 had misophonia. Misophonia co-existed with hyperacusis in 35 children (97%), and in one child misophonia occurred without hyperacusis. Misophonia was prevalent in older children, in females, and those with tinnitus. Fear and being upset were predominant emotional response in hyperacusis (without misophonia) while disgust and verbal abuse were prevalent in misophonia (with or without hyperacusis). Compared to children without DST, the hyperacusis (without misophonia) and misophonia (with or without hyperacusis) groups had significant higher prevalence of ADHD, anxiety, and language impairment. Educational difficulties were similar in APD irrespective of the presence or absence of DST. Despite higher tinnitus prevalence in misophonia (with or without hyperacusis) along with similar co-morbidities and educational difficulties in both hyperacusis (without misophonia) and misophonia (with or without hyperacusis), the misophonia (with or without hyperacusis) group surprisingly had less support at school which was reflected in fewer Education, Health and Care Plan (EHCP).

Conclusions: In APD misophonia mostly co-exists with hyperacusis, with differences in emotional responses, tinnitus prevalence, and gender distribution when compared to hyperacusis (without misophonia). Increase in awareness about misophonia is needed, as children with misophonia may have unidentified needs. Larger scale prospective study is required to clarify if misophonia evolves from hyperacusis, and to explore the factors underlying 'misophonia with hyperacusis' and 'misophonia without hyperacusis'. For clarity, DST studies need to specify if hyperacusis or misophonia co-existed when referring to hyperacusis or misophonia.

Key words: Hyperacusis, Misophonia, Decreased sound tolerance, Research Domain Criteria, Auditory Processing Disorder

Highlights:

- Research Domain Criteria frame-work is helpful in comprehensive assessment of children with auditory processing disorder (APD) and decreased sound tolerance (DST).
- First known study to explore prevalence of hyperacusis and misophonia in children with APD
- Misophonia in approximately 97% co-exist with hyperacusis
- Tinnitus is more common in misophonia (with or without hyperacusis) compared to hyperacusis (without misophonia)
- Predominant emotional response in hyperacusis (without misophonia) is 'fear' compared to 'verbal aggressiveness' in misophonia (with or without hyperacusis)
- Misophonia with hyperacusis is more common in females than hyperacusis (without misophonia)
- Despite significantly more co-morbidities compared to children with APD and hyperacusis (without misophonia), children with misophonia (with or without hyperacusis) are less likely to receive Education, Health and Care Plan.

1. Introduction

Children with oversensitivity to sounds or decreased sound tolerance (DST), that mainly relates to hyperacusis and misophonia, makes up a significant proportion of referrals to paediatric audiology clinics. Differentiating between hyperacusis and misophonia is important for appropriate management, which has been difficult due to lack of consensus on the definition, similarities in presentation, and descriptions that are based on diverse populations in different studies [1-4]. Hyperacusis and misophonia not only co-exist with each other [1] but also with other conditions. Overlapping co-morbidities is a recognised feature of neurodiverse and mental health conditions [5]. A diagnostic approach involving detailed clinical history [6] and evaluation of symptoms related to functional connectivity of related neural networks in identifying unique characteristics as well as features common to other co-morbid conditions in a specific population would be helpful. The Research Domain Criteria (RDoC) frame work is such an approach that has been recommended to be utilized in evaluating DST [2].

Swedo and colleagues (2022) suggested that misophonia is associated with impaired auditory processing, emotional regulation, and learning [3]. Association between auditory processing disorder (APD) and hyperacusis has been reported, both in children [7] and adults [8]. Not much is known about misophonia in APD. RDoC framework is already in clinical use to evaluate APD [9,10] which enquires about different aspects of DST in addition to relevant co-morbidities and educational concerns.

Current definitions of hyperacusis and misophonia lack clarity [1]. Hyperacusis is seen as DST related to the physical characteristics of sound such as loudness, but misophonia has also been mentioned to be partially related to physical characteristics of sounds. Both misophonia and hyperacusis are associated with strong emotional responses, arising from primary or secondary activation of the limbic system. Additionally, hyperacusis and misophonia have been reported to co-exist [1] but the extent of overlap is not known. Therefore it is not clear how to differentiate between hyperacusis and misophonia based on the physical characteristics of sound or specific emotional responses. To ensure clarity, the current study simply defined misophonia as over-sensitivity to sound of eating/chewing, the predominant trigger [11-13], and hyperacusis as oversensitivity to other sounds. Such definitions were felt as a good starting point to develop our understanding of misophonia and hyperacusis, with the scope to add additional characteristics based on the outcome of initial comparison. Based on data from routine APD clinical practice, this article aimed to explore:

i. Prevalence of DST in children with APD
ii. Extent of overlap between hyperacusis and misophonia
iii. Differentiating between hyperacusis (without misophonia) and misophonia (with or without hyperacusis) in the following areas:
    a. Age
    b. gender
    c. Different sound triggers
    d. Different emotional responses
    e. Co-morbidities

2. Material and methods

2.1. Study design

This study was an approved quality improvement project at a regional teaching hospital located in the Northwest region of the United Kingdom. Data were obtained retrospectively from health care records of children diagnosed with Auditory Processing Disorder (APD) in the paediatric audio-vestibular medicine department, between January 2021 and December 2022.

2.2. Participants

Two hundred and seventy-nine children (Males= 160, Females=119) with a diagnosis of APD, aged between 6 and 16 years (Mean =11.6 years; SD= 2.1), with a nonverbal intelligent quotient (NVIQ) ranging between 80-128 (Mean = 98.9; SD = 9.9) were studied. All participants were healthy English-speaking children attending mainstream schools. All the participants had pure tone audiometric thresholds ≤ 20 dB HL at 0.5, 1, 2, 4, 8, 10 and 12.5 kHz in both ears.

2.2.1. Hearing threshold

Pure-tone audiometry was carried out using a PC controlled AURICLE Aud (GN Otometrics) audiometer, operated from the OTOsuite Audiometry Module PC software, and Sennheiser HDA 300 headphones.

2.2.2. Tympanometry

GSI Graston Stadler TympStar Pro was used for tympanometry. Participants had middle ear pressures between +50 to -200 daPa.

2.2.3. NVIQ was evaluated using the fourth edition of the test for nonverbal intelligence [14]. Participants in this study had NVIQ ≥ 80, to ensure that the listening difficulty related to APD was not due to poor cognitive ability.

2.2.4. APD diagnosis

The APD diagnostic procedure involved structured history, physical examination, screening for common neurodevelopmental conditions, and diagnostic APD test battery. For detail of APD diagnosis please see Ahmmed et al. (2022).

2.3. Decreased sound tolerance and types

The structured history enquired about over-sensitivity to sounds, with four response choices: 'Never', 'Occasionally', Most times' and 'Always'. There were 37 various day-to-day sound choices in addition to an option of specifying other sounds not included in the questionnaire. Responses of 'Always' or 'Most times' were considered as significant DST, and 'Never' or 'Occasionally' were considered as 'No DST'. Impact of DST on the daily activities were assessed by responses: "Not sure", "No restriction", "Little (mild) impact", "Moderate", and "Severe". Participants were divided in three groups:

Group A- No DST

Group B- Hyperacusis (without misophonia): Oversensitivity to sounds other than eating/chewing.

Group C- Misophonia (with or without hyperacusis): Oversensitive to sounds of eating/chewing, with or without sensitivity to other sounds in Group B. The reason for not dividing misophonia into two groups, 'misophonia with hyperacusis' and 'misophonia without hyperacusis' is outlined in results section 3.1.

2.4. Other sensory over-sensitivities

Over-sensitivities to touch, smell, taste, eating, light, and pain were recorded.

2.5. Emotional responses

Different emotional responses triggered by the offending sounds were: being annoyed, anxious, complaining of loudness, crying/screaming, disgusted, distressed, frightened, gets angry, hits others, in pain, upset, and verbally abusive.

2.6. Co-morbidities

Common co-morbidities were assessed using validated questionnaires [15,16] that included:

2.6.1. Tinnitus

The presence or absence of tinnitus were noted in the history questionnaire.

2.6.2. Language Impairment (LI)

Children's Communication Checklist–2 [17] identified language impairment (LI) based on the General Communication Composite (GCC) and the Social Interaction Deviance Composite (SIDC). GCC <45 and SIDC ≤-15 were considered as LI.

2.6.3. Attention Deficit Hyperactivity Disorder (ADHD)

Strength and weakness in ADHD symptoms and normal behaviour (SWAN) rating scale was used to evaluate ADHD [18]. Inattention and impulsivity/hyperactivity symptoms were evaluated using nine items each. Averaged scores >2.11, >2.48, and >2 suggested combined, inattention and hyperactive types of ADHD respectively.

2.6.4. Manual dexterity

Fifth percentile cut-off in the manual dexterity sub-set of the second edition of the Movement Assessment Battery for Children (MABC-2) [19] was used to identify impaired manual dexterity.

2.6.5. Anxiety

The 24-item 'Anxiety Scale for Children with Autism Spectrum Disorder (ASC-ASD)' completed by children and carers evaluated anxiety [20]. The maximum total score is 72. Scores ≥ 20 suggested significant anxiety.

2.6.6. Academic concerns and support at school

The participants reading, spelling, writing and numeracy skills were recorded as 'very good', 'good', 'average', 'poor', or 'very poor' by the parents/carers. 'Very poor' and 'poor' were considered as educational concern. Participants who had Education, Health and Care Plan (EHCP) at school were recorded.

2.7. Data analysis

SPSS version 29 was used for statistical analyses. The data were not normally distributed. Kruskal-Wallis tests carried out for continuous variables. Bonferroni correction of p value were carried out for multiple tests, and values <.05 were considered as statistically significant. Cross tabulations and Chi-square tests were carried out for categorical data.

3. Results

3.1. Prevalence and types of DST in children with APD

One hundred and thirty six participants (48.7%; 95% CI 42.7%-54.8%) did not have DST (Group A), 107 (38.4%; 95% CI 32.6%-44.3%) had hyperacusis without misophonia (Group B, over-sensitive to sounds other than eating/chewing), and 36 (12.9%; 95% CI 9.2%-17.4%) had misophonia (Group C, over-sensitive to the sound of eating/chewing, with or without hyperacusis). Only a 12.5 year old male in Group C with NVIQ of 109, anxiety, impaired manual dexterity, language impairment, and oversensitivity to touch sensation developed misophonia only at the age of 10 years without any symptoms of hyperacusis. Due to the single case of misophonia without hyperacusis, all participants with oversensitivity to sound of eating/chewing irrespective of the presence or absence of hyperacusis were considered together in Group C as misophonia.

3.2. Age, gender and NVIQ

Group A comprised 82 males and 54 females, 6 - 16 year-olds (mean 11.7, SD 2.1), NVIQ 81-125 (mean 98.7, SD 9.6). Group B comprised 66 males and 41 females, 7-16 year-olds (mean 11.3, SD 2.1), NVIQ 80-127 (mean 99.5, SD 10.1). Group C comprised 12 males and 24 females, 7-16 year-olds (mean 12.6, SD 2.4), NVIQ 84-128 (mean 97.3, SD 10.6). Significantly more females than males were notes in Group C, compared to groups A [χ^2 (1, N=172) =8.35, p<.01] and B [χ^2 (1, N=143)= 8.73, p<.01]. Gender differences between Groups A and B were not significant, with significantly more males in Groups A and B compared to Group C. Participants in group C were significantly older than groups A and B (p<.01). NVIQ were not different between the groups (p>.05).

3.3. Different sound triggers

The number of sound triggers varied, ranging from one to 28 sounds (average 12) in group B, and from one to 36 sounds (average 14) sounds in addition to sound of eating in group C. In group C the oversensitivity to the sole sound was the 'sound of eating/chewing' and was noted in only one child. Detailed comparison between the groups is shown in Table 1. Some triggers were common in both groups such as 'unexpected sounds', 'any crowed places', 'loud voices', 'hand-dryers', 'firework' amongst others. Significantly larger numbers of group C participants were oversensitive to different body sounds other than eating/chewing, tapping, running taps and playground noises than those in group B.

Table 1 Here

3.4. Emotional/psychological responses and impact on daily activity

Table 2 compares emotional responses between groups B and C. Being "upset" and "frightened" were common in Group B ($p<.05$). Verbal abuse and feeling of disgust were common in Group C ($p<.01$). Figure 1 shows that complaint of loudness, annoyance, fear, and pain overlapped.

Table 2. Here

Figure 1. Here

Sixty-three percent children in group B and 58.3% in group C reacted on anticipation of the trigger sounds ($p>.05$). In group B, 33 of the 107 (30.8%) participants and in group C, five of the 36 (13.8%) participants used ear defenders [χ^2 (1, N=143)=3.967; $p<.05$).

3.5. Comorbidities

3.5.1. Other perceptual over-sensitivities

Participants in the groups B and C had other sensory issues, detailed in table 3. No statistically significant differences were seen between the two groups in terms of over-sensitivity to touch, smell, taste, eating, pain and light between the two groups ($p>.05$).

Table 3. Here

3.5.2. Tinnitus

Compared to Group A, significantly more participants reported tinnitus in groups B [χ^2 (1, N=237)=4.898; $p<.05$] and C [χ^2 (1, N=168)=15.272; $p<.0001$]. Significantly higher report of tinnitus was also seen in group C compared to group B [χ^2 (1, N=141)=4.465; $p<.05$].

3.5.3. Language impairment

LI was identified in 96 (70.5%), 97 (90.6%), and 32 (88.8%) children in groups A,B, and C respectively. The scatterplot for GGC and SIDC values is shown in figure 2. Comparison of the GCC scores showed significant difference between groups A, B and C [$H(2)=40.775$; $p<.001$]. Compared to group A, both groups B ($p<.001$) and C ($p<.01$) had low GCC. SIDC helps to identify the type of LI, either structural (SIDC >8), or Pragmatic (SIDC <0), or intermediate to indicate either structural or pragmatic impairment or a combination of both. Structural plus intermediate categories have higher probability of structural LI (SLI), whereas pragmatic plus intermediate categories have a higher probability of pragmatic impairment (PLI). There were 49, 47 and 13 participants within the SLI category in groups A, B, and C respectively [χ^2 (2, N=279)=1.720; $p>.05$]. There were 80, 88 and 29 participants within the PLI category in groups A, B, and C respectively [χ^2 (2, N=279)=17.797; p<.001]. Pairwise comparison showed significantly larger number of PLI in group B compared to group A [χ^2 (1, N=243)=14.754; p<.001], and in group C compared to group A [χ^2 (1, N=143)=5.791; p<.05]. No differences was noted between groups B and C.

Figure 2 Here Please

3.5.4. ADHD symptoms

SWAN rating score were missing for one participant in group A, for 2 participants in group B and one participant in group C. Based on 5th centile cut-off for SWAN score, eight participants in group A (6%), 20 in group B (19%), and eight in group C (23%) had significant ADHD symptoms, which was significant [χ^2 (2, N=275)=12.300; p<.01]. Compared to group A, more ADHD were diagnosed in groups B [χ^2 (1, N=240)=9.867; p<.01] and C [χ^2 (1, N=170)=9.345; p<.01]. No difference noted between groups B and C (p>.05).

3.5.5.Manual dexterity

Using the 5th percentile cut-off in the manual dexterity sub-set of MABC-2, impaired manual dexterity was identified in 66 (48.5%), 53 (49.5%), and 12 (33.3%) of participants in groups A,B, and C respectively. No significant differences were found between the groups (p>.05).

3.5.6. Anxiety

The Anxiety Scale for Children-ASD (ASC-ASD) questionnaire completed by parents identified anxiety issues in 53 (39%), 80 (75%), and 28 (78%) of children in groups A, B, and C, which was significant [χ^2 (2, N=278)=39.257; p<.00001]. There was one missing data for group B. Compared to group A, larger number of children had significant anxiety in both groups B [χ^2

(1, N=242)=32.062; p<.0001]and C [χ^2 (1, N=172)= 17.205; p<.0001]. No significant difference was noted between groups B and C (P>.05).

The questionnaire completed by the children identified anxiety in 62 (46.6%), 73 (71.5%) and 30 (83.3%) of children in groups A, B, and C respectively [χ^2 (2, N=271)=23.876; p<.00001]. Compared to Group A, more participants had significant anxiety scores in both groups B [χ^2 (1, N=235)=14.702; p<.001] and C [χ^2 (1, N=169)=15.339; p<.001].

3.5.7. Overlapping co-morbidities

Figures 3a and 3b demonstrate overlapping co-morbidities in the misophonia and hyperacusis groups respectively. ADHD symptoms were only seen in those who also had language impairment.

Figure 3a Here

Figure 3b Here

3.5.8. Educational difficulties

Parents raised concerns about reading in 46 (33.8%), 45 (42%), and 10 (27.7%) children in groups A, B, and C respectively [χ^2 (2, N=279)=3.027; p>.05]. Concerns about spelling ability were raised in 69 (50.7%), 57 (53.2%), and 19 (52.7%) children in groups A, B, and C respectively [χ^2 (2, N=279)=0.165; p>.05]. Poor or very poor numeracy skills were reported in 58 (42.6%), 48 (45.2%), and 15 (41.6%) children in groups A, B, and C respectively [χ^2 (2, N=278)=0.226; p>.05]. There was one missing data for group B.

3.5.9. *Education and Health Care Plan (EHCP)*

30 (22.5%), 38 (36.8%), and 6 (16.6%) children in groups A, B, and C respectively had EHCP, and the group differences was significant [$\chi^2(2,N=272)=8.351$; $p<.05$]. There were few missing data, 3 in group A and 4 in group B. Significantly more participants in group B had EHCP compared to groups A [χ^2 (1, N=236)=5.816; $p<.05$] and C [χ^2 (1, N=139)=5.044; $p<.05$].

4. Discussion

4.1. Sound triggers

The findings in this study of significant over-sensitivity to tapping, clicking and different body sounds other than eating and chewing in misophonia (with or without hyperacusis) is consistent with the literature (Table 1). The trigger of tapping and clicking sound in 63% of participants with misophonia in this study is not dissimilar to 74%, 54%, 38.4%, 25.4% and 14% reported by Brennan et al. (2024), Guzick et al. (2022), Jager et al. (2020); Rosenthal et al. (2022) and Siepsiak et al. (2023) respectively [12, 21-24]. Breathing sounds as a trigger in 55.5% cases in this study also compares well with 59% reported by [24]. Our study adds that sounds of running water taps and noises in the playground are also misophonia triggers.

Over-sensitivity to sounds such as unexpected sounds, noises associated with a crowd in school or social gathering, loud voices, sirens/alarms, fireworks amongst others present in both hyperacusis and misophonia is consistent with the view that misophonia coexists with hyperacusis [1, 23]. Our study adds to the literature that in about 97% cases misophonia co-existed with hyperacusis (35 out of 36 children with misophonia had hyperacusis, see Results section 3.1).

4.2. Emotional responses

Tyler et al. (2014) subcategorised hyperacusis as loudness, annoyance (misophonia) fear (phonophobia) and pain hyperacusis[25]. The present study suggests loudness, annoyance, fear and pain responses co-exist (Table 2, and Figure 1) in hyperacusis (without misophonia) and misophonia (with or without hyperacusis), consistent with Brennan et al. (2024) [23]. There were no differences in complaint about loudness between hyperacusis (without misophonia) and misophonia (with or without hyperacusis), and distinction between them based on oversensitivity to physical characteristics of sounds, suggested by Jastreboff and Jastreboff [1], is difficult. Consistent with Tyler et al. (2014) 'fear' was the predominant response in hyperacusis (without misophonia) [25], in addition to 'being upset' by sounds. The low prevalence of fear in misophonia (with or without hyperacusis) noted in this study is consistent with Siepsiak et al. (2023)[24].

Verbal abuse and being disgusted by the trigger sounds were significantly high in misophonia (with or without hyperacusis) in this study. Disgust has been reported in misophonia in 57% by Siepsiak et al. (2023) [24], 64% by Jager et al. (2020)[12], and 68% by Guzick et al. (2022) [21]. High levels of verbal aggression in misophonia has been reported in the literature [21].

4.3. Co-morbidities

The overlap of multiple co-morbidities in this study in hyperacusis (without misophonia) (Figure 3a) and misophonia (with or without hyperacusis) [Figure 3b] is consistent with the RDoC framework [5] and APD literature [9].

4.3.1 Language impairment

More than 90% of participants in the hyperacusis (without misophonia) and misophonia (with or without hyperacusis) groups in this study had language impairment, mainly pragmatic language impairment (Figures 2, 3a & 3b). Previous studies have demonstrated high levels of pragmatic language impairment in children with DST in APD but no distinction was made between hyperacusis and misophonia [7]. To the best of the authors knowledge, this is the first study attempting to distinguish hyperacusis and misophonia in children with APD. This study suggests that hyperacusis and misophonia could not be differentiated based on the language impairment, using CCC2 outcome in children with APD.

4.3.2. Anxiety

More than 70 % participants in both the hyperacusis (without misophonia) and misophonia (with or without hyperacusis) groups had anxiety issues, significantly more than those without DST. The above finding is consistent with the literature in hyperacusis [26], misophonia [27] and sensory over-responsivity [28]. Siepsiak et al. (2022) reported high anxiety levels in misophonia, but comparison with hyperacusis was difficult due to the small number of participants in the hyperacusis group [29]. Guzick et al. (2022) reported social anxiety disorder and generalized anxiety disorder in 30% and 27% of youth with misophonia respectively [21], and Rosenthal et al. (2022) reported 56.9% of individuals with misophonia met the full criteria for at least one anxiety disorder [22]. Rinaldi et al. (2022) also showed significant anxiety levels in children with misophonia [30]. However, unlike the current study, the above studies did not compare anxiety between misophonia and hyperacusis. Anxiety score was unable to differentiate between hyperacusis (without misophonia) and misophonia (with or without hyperacusis).

4.3.3. ADHD

ADHD identified in this study in APD is consistent with the literature [31], so is the association with misophonia. Twenty-three percent of participants in this study diagnosed with ADHD in the misophonia (with or without hyperacusis) group is consistent with 21% and 17.9% ADHD reported in misophonia by Guzick et al.(2022) and Rosenthal et al. (2022) respectively [21, 22]. Our study adds to the literature by demonstrating, firstly the presence of ADHD did not differentiate between hyperacusis (without misophonia) and misophonia (with or without hyperacusis); secondly presence of hyperacusis or misophonia increases the possibility ADHD in APD; and thirdly ADHD in hyperacusis and misophonia is present in association with language impairment (see Figures 3a and 3b).

4.3.4. Impaired Manual Dexterity

The recognition of the importance of sensory processing and motor function in mental health resulted in the inclusion of sensori-motor system in RDoC [32]. Impaired motor function is seen in many neurodevelopmental conditions associated with misophonia and hyperacusis, such as ADHD [33], APD [16], and ASD [34]. The current study adds to the literature by showing that impaired manual dexterity in APD was not influenced by DST.

4.3.5. Tinnitus

Hyperacusis has been suggested as a precursor of tinnitus [35], and the current study show that tinnitus is more related to misophonia (with or without hyperacusis). The higher proportion of participants with misophonia to have tinnitus compared to the hyperacusis (without misophonia) and no DST groups in the current study is consistent with Brennan et al. (2024) [23] and Aazh et al. (2022) [27]. Brennan et al. (2024) reported significant association between misophonia and tinnitus [23]. Report of tinnitus in 74% of individuals with misophonia by Aazh et al. (2022) [27] is consistent with our study findings of 50 % children with APD and misophonia (with or without hyperacusis) to have tinnitus compared to 30.5% in the hyperacusis (without misophonia) and 18.2% in the no DST groups.

4.3.6. Other sensory oversensitivity

Not many studies compared other sensory issues in misophonia and hyperacusis. Rosenthal et al. (2022) reported sensory processing difficulties in 9.7% of misophonia participants, but no comparison was made with hyperacusis [22]. Our study shows that other sensory over-sensitivities varied from 22% to 58% depending on the sensory domain and there were no significant differences between hyperacusis (without misophonia) and misophonia (with or without hyperacusis) [Table 3].

4.4. Age

Participants with misophonia (with or without hyperacusis) with mean age of 12.6 years were significantly older than hyperacusis (without misophonia) in this study, consistent with the mean ages of 11.7 and 13.7 years reported in misophonia by Rinaldi et al. (2022) and Guzick et al. (2022) respectively [21, 30]. However, the above two studies did not have a hyperacusis comparison group.

Our current study suggests DST is first noted in infancy, but it is unclear about the initial sounds they were sensitive to. Siepsiak et al. (2023) reported that in 4 out of 45 children with misophonia oversensitivity to sounds of eating was present by the age of 3 years, but there was no hyperacusis group to compare with [24].

4.5. Gender

The high prevalence of misophonia (with or without hyperacusis) in females (67%) in this study is consistent with Rinaldi et al. (2022), Guzick et al. (2022), Jager et al. (2020), and Rosenthal et al. (2022) reporting 60%, 68%, 70.9% and 74.4% female prevalences of misophonia respectively [12, 21, 22, 30]. Our studies adds to the literature by demonstrating higher female prevalence of misophonia (with or without hyperacusis) compared to hyperacusis (without misophonia). This is interesting as both hyperacusis and misophonia co-exists with neurodevelopmental conditions like ADHD, APD and ASD [7, 21, 22, 36, 37] that are more prevalent in males [38, 39]. In the current study, in terms of co-morbidities misophonia (with or without hyperacusis) group had higher prevalence of tinnitus compared to the hyperacusis (without misophonia) group, and no significant differences between the groups in respect of ADHD, anxiety and LI (see Results). Rinaldi et al., (2022) suggest that misophonia is associated with poorer well-being [30]. It was therefore, surprising to see that these children with misophonia (with or without hyperacusis) had fewer EHCP for support at school. Hyperacusis may be seen as a precursor of misophonia and the same neural system is involved in both the conditions [1]. Emotional processing and different expressions of mental disorders are related to differential hypo- and hyper-activation of neural networks within the same system [40] and it is possible that anxiety and stress [41] related to unmet needs may be the trigger for misophonia and other associated mental health issues [22] or non-organic hearing loss [42] in children with APD. Another reason for lower EHCP in misophonia could be due to females internalizing the symptoms [43] making identification by professionals delayed and difficult. Moreover, it could be related to lack of awareness of association between mental health and sensory oversensitivity [32]. Higher prevalence of certain mental health issues in females [44] and difficulties accessing Child and Adolescent Mental Health Services (CAMHS) could be another factor [45]. Future research is need to answer the underlying reason for the higher predominance of female in misophonia (with or without hyperacusis), and to compare the functional activities within the neural networks in hyperacusis and misophonia. In view of co-morbidities it is important that children with APD are evaluated holistically with a neurodevelopmental pathway similar to ADHD and ASD.

4.6. Prevalence

No other studies have compared the prevalence of hyperacusis and misophonia in APD, therefore, we can also compare the findings of this study with prevalence of hyperacusis and misophonia in some co-morbid conditions. The 38.4% (95% CI 32.6%-44.3%) prevalence of hyperacusis (without misophonia) in APD in this study is consistent with 37-45% prevalence of hyperacusis reported in autism spectrum disorder (ASD) [37]. There is no consensus and the prevalence of misophonia varies with diagnostic criteria used [23]. The 35.5% prevalence of misophonia in ASD [46] using the Duke-Vanderbilt Misophonia Questionnaire (DVMSQ) was higher than 12.9% (95% CI 9.2%-17.4%) found in this study from clinical interview and structured history questionnaire in a routine APD clinic. However, 36 participants with misophonia (with or without hyperacusis) out of 143 participants with DST (25%) in the current study is consistent with report of misophonia in 23% of patients attending hyperacusis and tinnitus clinics [27]. Our study adds to the literature that misophonia co-exists with hyperacusis in approximately 97% cases (in 35 out of 36 children with APD).

4.7. Limitation of the study

The current study, being a retrospective study of 6-16 year olds, was unable to explore the progress of the hyperacusis and misophonia from the onset. The study found that only one out of 36 participants with misophonia did not have hyperacusis. Therefore, this study was unable to explore features of 'misophonia without hyperacusis' and larger studies are needed. Additionally, this study evaluated only a limited number of constructs, sub-constructs and systems of the RDoC framework. Future studies are required with more multidisciplinary input exploring other relevant RDoC components, such as visual processing. It would also be worth future studies to explore any distinguishing features between individuals with 'oversensitivity to sound of eating/chewing' versus 'oversensitivity to other body generated sounds and repetitive tapping sounds without oversensitivity to sound of eating/chewing'.

5. Conclusion

This study compared hyperacusis (oversensitivity to sounds other than eating/chewing), misophonia (oversensitivity to sounds of eating/chewing with or without hyperacusis) and no decreased sound tolerance (no DST) groups in children diagnosed with APD using RDoC framework. Fifty-one percent had DST, constituting 75% hyperacusis and 25% misophonia. Misophonia mostly co-existed with hyperacusis. Misophonia (with or without hyperacusis) was prevalent in females and in older children compared to hyperacusis (without misophonia). Compared to those with APD without DST, both hyperacusis (without misophonia) and misophonia (with or without hyperacusis) were associated with ADHD, anxiety, educational difficulties, and LI. In addition to oversensitivity to eating/chewing sounds, children with misophonia plus hyperacusis had higher prevalence of oversensitivity to other body sounds, repetitive clicking/tapping, running taps and playground noises.

Issues with loudness, annoyance, fear and pain overlapped, with fear and being upset with sounds being prevalent responses in hyperacusis (without misophonia), while prevalent responses in misophonia (with or without hyperacusis) were verbal abuse and disgust. Despite similar comorbidities and higher prevalence of tinnitus compared to children with APD plus hyperacusis (without misophonia) and higher prevalence of different co-morbidities including tinnitus compared to children with APD without DST, children with APD and misophonia (with or without hyperacusis) had fewer EHCP compared to the other two groups. The need for increased vigilance of DST in females is suggested to ensure they do not have any unmet needs. The importance of holistic evaluation of children with DST and APD within established neurodevelopmental pathway used for ADHD and ASD is highlighted. For clarity, DST studies need to specify if hyperacusis or misophonia co-existed when referring to hyperacusis or misophonia.

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Table 1. Comparison of sound triggers between hyperacusis (without misophonia) and misophonia (with or without hyperacusis).

Trigger Sounds	**Group B: Hyperacusis N (%) DST to trigger**	**Group C: Misophonia N (%) DST to trigger**	**χ^2 (DST Group B vs. C)**
Eating	0	36 (100%)	
Unexpected sounds	71 (66.3%)	25 (69.4%)	χ^2 (1, N=143) = 0.116; p = .732
Any crowded place	61 (57%)	24 (66.6%)	χ^2 (1, N=143) = 1.042; p = .307
Classroom	61 (57%)	24 (66.6%)	χ^2 (1, N=143) = 1.042; p = .307
Loud voice	72 (67.2%)	24 (66.6%)	χ^2 (1, N=143) = 0.004; p = .945
Tapping /clicking	**20 (18.6%)**	**23(63.8%)**	**χ^2 (1, N=143)= 26.168; p<.0001***
School dining room	53 (49.5%)	22 (61.1%)	χ^2 (1, N=143) = 1.447; p = .228
Breathing	**10 (9.3%)**	**20 (55.5%)**	**χ^2 (1, N=143)= 34.696; p<.0001***
Drills	59 (55.1%)	19 (52.7%)	χ^2 (1, N=143) = 0.060; p = .805
Hand Dryer	53 (49.5%)	18 (50%)	χ^2 (1, N=143) = 0.002; p = .961
Firework	66 (61.6%)	17 (47%)	χ^2 (1, N=143) = 2.312; p = .128
Coughing	**9 (8.4%)**	**17 (47.2%)**	**χ^2 (1, N=143)= 27.275; p<.0001***
Siren	56 (52.3%)	16 (44.4%)	χ^2 (1, N=143) = 0.671; p = .412
School assembly	39 (36.4%)	16 (44.4%)	χ^2 (1, N=143) = 0.727; p = .393
Crying	33 (30.8%)	16 (44.4%)	χ^2 (1, N=143) = 2.213; p = .136
Balloon Popping	58 (54.2%)	15 (41.6%)	χ^2 (1, N=143) = 1.69; p = .192
Motor Bike	46 (42.9%)	15 (41.6%)	χ^2 (1, N=143) = 0.019; p = .889
Disco/Party	55 (51.4%)	15 (41.6%)	χ^2 (1, N=143) = 1.021; p = .312
Playground	**20 (18.7%)**	**15 (41.6%)**	**χ^2 (1, N=143) =7.520; p< .01***
Hoover	41 (38.3%)	15 (41.6%)	χ^2 (1, N=143) =0.126; p = .721
Sneezing/sniffing	**9 (8.4%)**	**14 (38.8%)**	**χ^2 (1, N=143) = 18.538 p<.0001***
Lawn mover	29(27.1%)	13 (36.1%)	χ^2 (1, N=143) = 1.0538; p =.3046
Hair Dryer	33 (30.8%)	11 (30.5%)	χ^2 (1, N=143) = .001; p = .9743
Whistle	30 (28%)	11 (30.5%)	χ^2 (1, N=143) = 0.0835; p = .7725
Restaurant	19 (17.7%)	11 (30.5%)	χ^2 (1, N=143) = 2.661; p = .102
General Traffic	25 (23.4%)	10 (27.7%)	χ^2 (1, N=143)0.2838; p = 0.594
Tap Running	**5 (4.6%)**	**9 (25%)**	**χ^2 (1, N=143) =12.602; p <.001***
Thunder	45 (42 %)	9 (25%)	χ^2 (1, N=143) = 3.33; p = .67
Supermarket	30 (28%)	9 (25%)	χ^2 (1, N=143) = 0.125; p = .723
Bell	40 (37.4%)	9 (25%)	χ^2 (1, N=143) = 1.833; p = .175
Musical Instrument	26 (24.2%)	7 (19.4%)	χ^2 (1, N=143) = 0.357; p = .549
Fan	6 (5.6%)	5 (13.8%)	χ^2 (1, N=143) = 2.601; p = .106
Animals	10 (9.3%)	5 (13.8%)	χ^2 (1, N=143) = 0.592; p = .441
Cinema	21 (19.6%)	5 (13.8%)	χ^2 (1, N=143) = 0.596; p = .440
Washing Machine	8 (7.5%)	5 (13.8%)	χ^2 (1, N=143) = 1.340; p = .247
Birds	7 (6.5%)	3 (8.3%)	χ^2 (1, N=143) = 0.132; p = .715
Rain	5 (4.6%)	2 (5.5%)	χ^2 (1, N=143) = 0.045; p = .831

** Significant group differences in bold.*

Table 2. Comparison of emotional responses between hyperacusis (without misophonia) and misophonia (with or without hyperacusis).

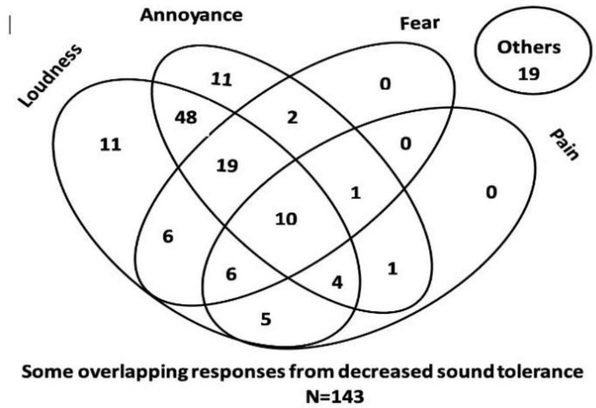
Responses	Group B:Hyperacusis N (%) with responses	Group C:Misophonia N (%) with responses	Chi-Square statistics
Annoyed	68(64%)	29(80%)	χ^2 (1, N=143)=3.569; p>.05
Loudness complaint	82(77%)	27(75%)	χ^2 (1, N=143)=0.039; p>.05
Distressed	68(64%)	22(61%)	χ^2 (1, N=143)=0.068; p>.05
Anger	54(50%)	22(61%)	χ^2 (1, N=143)=1.225; p>.05
Anxious	65(61%)	17(47%)	χ^2 (1, N=143)=2.014; p>.05
Verbally abusive	**21(17%)**	**16(44%)**	**χ^2 (1, N=143)=8.650; p<.01***
Upset	**61(57%)**	**10(28%)**	**χ^2 (1, N=143)=9.207; p<.01***
Cries/screams	31(29%)	9(25%)	χ^2 (1, N=143)=0.210; p>.05
Disgusted	**6(6%)**	**8(22%)**	**χ^2 (1, N=143)=8.419; p<.01***
In pain	22(21%)	7(19%)	χ^2 (1, N=143)=0.020; p>.05
Hits own head	18(17%)	6(17%)	χ^2 (1, N=143)=0.001; p>.05
Hits others	11(10%)	3(8%)	χ^2 (1, N=143)=0.115; p>.05
Frightened	**44(41%)**	**3(8%)**	**χ^2 (1, N=143)=13.124; p<.001**

** Significant group differences in bold.*

Table 3. Other perceptual over-sensitivities in addition to decreased sound tolerance.

Perceptions	**Group B: Hyperacusis N (%) with sensitivity**	**Group C: Misophonia N (%) with sensitivity**	**Chi-Square statistics**
Touch	48(45%)	21(58%)	χ^2 (1, N=143)=1.958; p>.05
Fussy eating	58(54%)	15(42%)	χ^2 (1, N=143)=1.694; p>.05
Smell	51(48%)	13(36%)	χ^2 (1, N=143)=1.454; p>.05
Taste	42(39%)	10(28%)	χ^2 (1, N=143)=1.532; p>.05
Pain	33(31%)	9(25%)	χ^2 (1, N=143)=0.443; p>.05
Light	24(22%)	8(22%)	χ^2 (1, N=143)=0.001; p>.05

Figure 1. Overlap between loudness, annoyance, fear and pain responses in decreased sound tolerance.



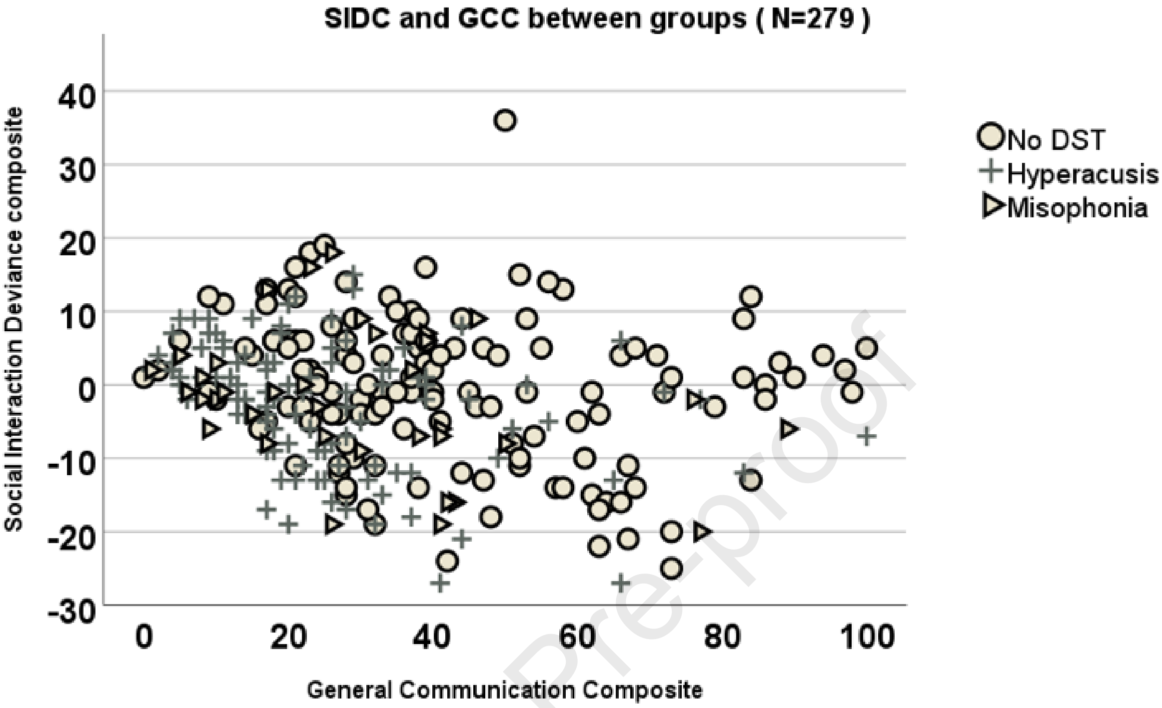


Figure 2. Scatterplot showing the distribution of general communication composite (GCC) and social interaction deviant composite(SIDC) in the No DST, hyperacusis (without misophonia) and Misophonia (with or without hyperacusis). GCC ≤ 45 =LI; SIDC ≥9= structural language impairment; SIDC 0-8= Intermediate group; SIDC <0= pragmatic language impairment.

Figure 3 a. **Overlap of language impairment, ADHD, anxiety and impaired manual dexterity in hyperacusis (without misophonia)**

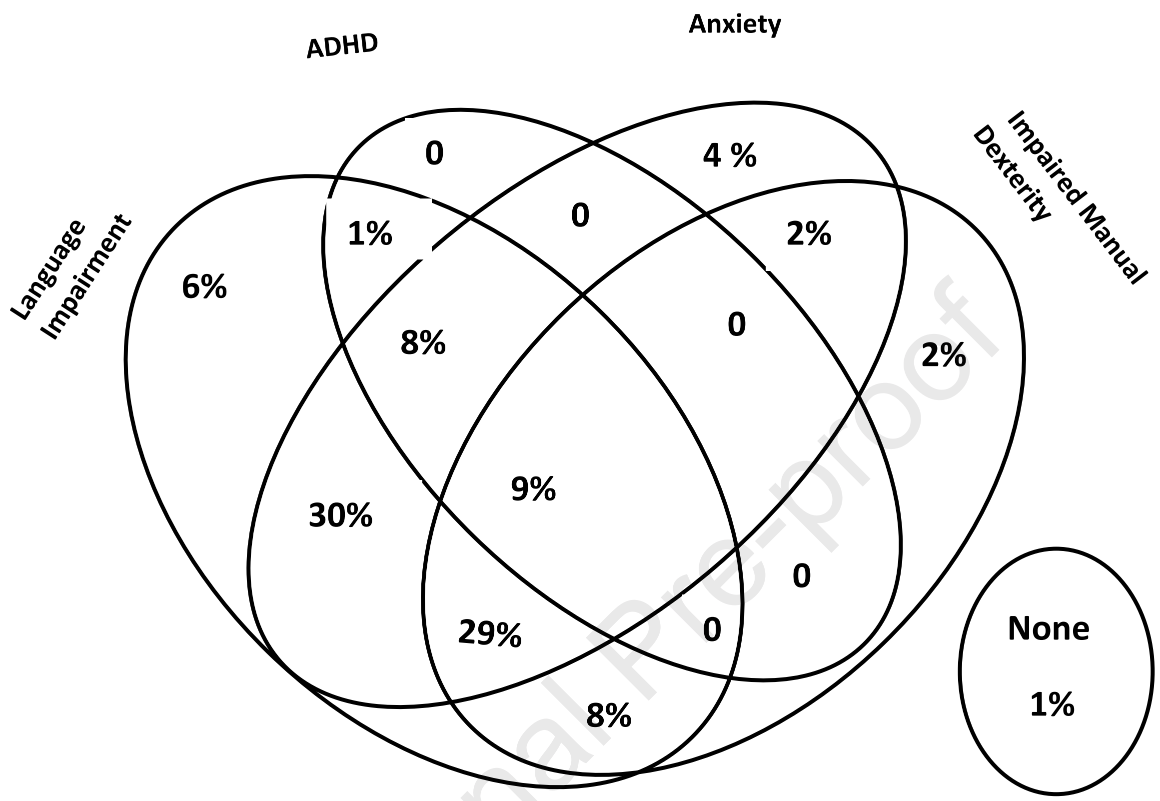
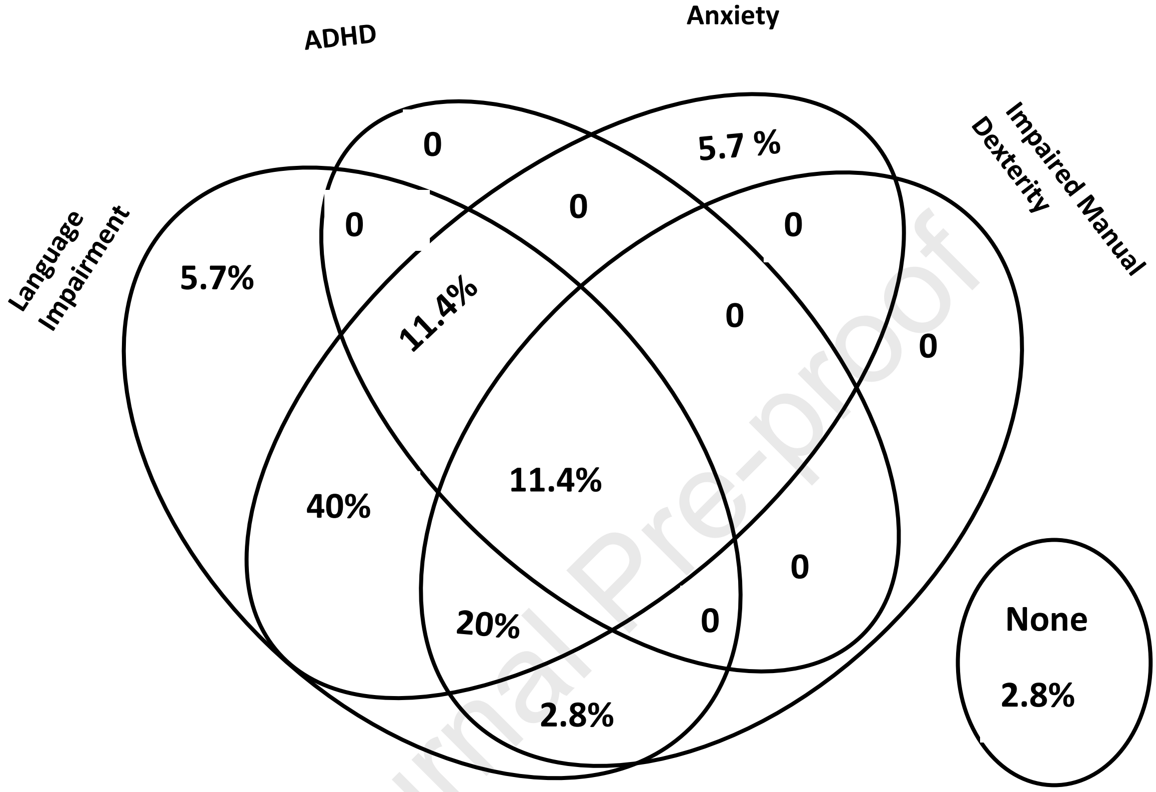


Figure 3b. **Overlap of language impairment, ADHD, anxiety and impaired manual dexterity in Misophonia (with or without hyperacusis)**



Highlights:

- **Research Domain Criteria frame-work is helpful in comprehensive assessment of children with APD and decreased sound tolerance**
- **First known study to explore prevalence of hyperacusis and misophonia in children with APD**
- **Misophonia in approximately 97% co-exist with hyperacusis**
- **Tinnitus is more common in misophonia (with or without hyperacusis) compared to hyperacusis (without misophonia)**
- **Predominant emotional response in hyperacusis (without misophonia) is 'fear' compared to 'verbal aggressiveness' in misophonia (with or without hyperacusis)**
- **Misophonia with hyperacusis is more common in females than hyperacusis (without misophonia)**
- **Despite significantly more co-morbidities compared to children with APD and hyperacusis (without misophonia), children with misophonia (with or without hyperacusis) are less likely to receive Education, Health and Care Plan**

Declaration of interests

☒ The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

☐ The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: